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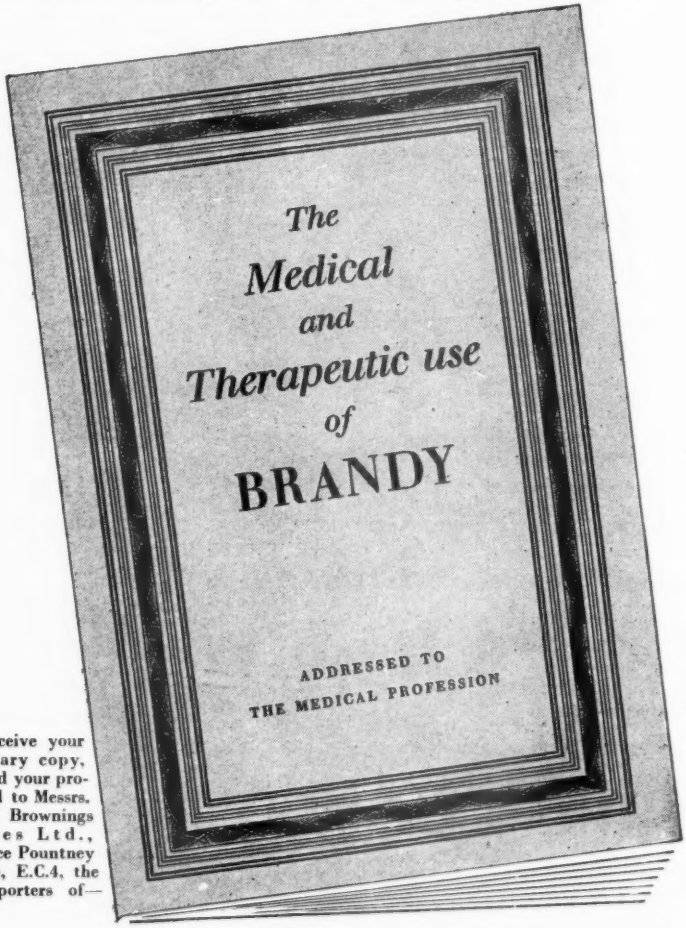

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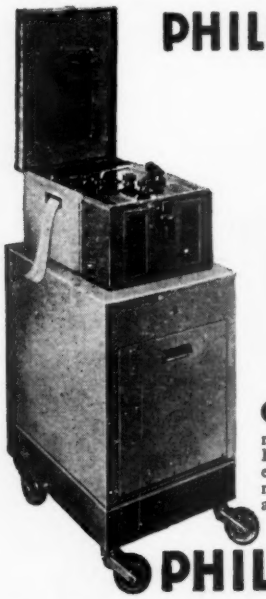

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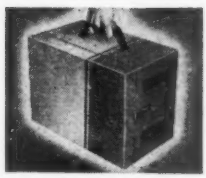

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Section of General Practice

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[*February* 17, 1954]

DISCUSSION: SOME ASPECTS OF PSYCHIATRY IN GENERAL PRACTICE

Dr. J. Whitby, London:

Some Personality Types as Seen in General Practice

The purpose of this paper is to point out some lessons learned as a psychiatrist which I have since found of value in the daily work of a general practitioner. By occasionally reviewing our dealings with our patients over a period of time we learn to know our patients. Certain personality types emerge that react to more or less slight psychological or physical stresses by episodes of illness. The problem here is recognizing and handling a personality rather than treating an illness. A common type is:

The anxious personality.—The usual reaction to environmental stress is by anxiety, depression or a mixture of both. Anyone can get an anxiety episode or a reactive depression with sufficient stress but the anxious personality gets repeated episodes with slight stresses. Although normally apprehensive or highly strung, they generally come to the doctor for somatic manifestations of anxiety, often multiple. Such are continuous headache or pressure on top of the head, dizziness, palpitation, abdominal symptoms or frequency. Sometimes they complain mostly of the more psychological symptoms such as insomnia, difficulty in concentration, irritability or feeling on edge, although such symptoms are always present if looked for. Recognition is easy especially if insomnia and mood are enquired after. The reply to some such question as "How do you feel in yourself, or in your spirits?" will be "Edgy" or "Depressed".

The patient who only comes for periodic exacerbations due to environmental stress is easily treated but not by saying "Pull yourself together" which excites shame or resentment, nor by saying "It's only your nerves" which leaves the patient with the feeling she has some mysterious disease. Sedatives are of value while the patient is recovering, but contribute nothing to the long-term handling of the case and prevention of future illness. What this requires is training the patient to talk about her difficulties and not her symptoms. The first step is to do a physical examination and reassure the patient by saying she is organically sound, but it is essential to go on and explain how the symptoms arise. One must relate the symptoms to common experience, e.g. the stomach turns over when you hear a bomb fall, diarrhœa or palpitation before an examination or interview, dizziness and blackouts due to blood rushing away from the head—the opposite of blushing. One emphasizes that the symptoms are real disturbances not imaginary ones, but are not due to local disease, only to a general state of tension. "Blushing is not a skin disease" is an obvious parallel. Spending five minutes on this explanation on the first occasion saves time, because it only has to be done once and one does not need to examine the patient any more. If it is omitted they need repeated examination and reassurance. These patients readily accept the idea that such symptoms can be produced by nervous strain, and then the rest gushes out. The patient who says "But I've no worries, doctor," means she thinks she ought not to be worried by trivialities, and if one tells her that the symptoms are evidence that she is more worried than she realizes, and casually adds some such remark as "Two children are enough worry for anyone" it generally sinks in by next time that she need not be ashamed to admit her difficulties. Men can always be approached through their work, and women through their home. One makes a note like dyspepsia—mother-in-law, or headaches—boy friend, and next time the patient comes in with such symptoms one starts at once "How's the mother-in-law or the boy friend behaving?" or "Have you still got the same forewoman at work?" One thus paves the way for what need only be quite short friendly chats with common-sense advice. Often all that is needed is to present the problem clearly to the patient who then finds her own solution, or alternative suggestions may be offered from which the patient may choose. Even if the problem is insoluble it helps the patient to get it off her chest. The doctor gradually enlarges his knowledge not only by experience, but by information which patients find out and which he notes in his address book, so that it becomes a directory of local social agencies which can help. Thus he may refer a patient in difficulties with her husband to the Probation Officer or Poor Man's Lawyer; the burden of the invalid father will be eased by co-operation with the geriatric unit, hospital almoner or the W.V.S. He will know how the personnel departments of factories will gladly meet him half-way if a patient is having difficulties at work, he will know the addresses of local clubs suitable for his adolescents, he will be able to help the unmarried mother through a moral welfare society. He will know what are the resources of the Invalid Children's Aid Association, the Red Cross and similar organizations. By handling patients in this way, we not only get to know each patient's difficulties and reactions, but the patient gets to understand how they cause her symptoms,

accepts the knowledge, comes less often for tonics and gradually adapts to her sensitiveness. There are two types of case that cannot be handled in this way. One is the agitated depression of psychotic or near psychotic severity which is diagnosed by noting that there is a marked change from the previous personality and then enquiring more closely into the mood. This needs early psychiatric treatment but experience shows that this is unfortunately often delayed through giving routine sedatives for somatic anxiety symptoms. The other is the elderly patient where age or arteriosclerosis has limited his power to adapt and he responds by agitation or depression to loss of spouse, change of residence, loss of a pet animal, or merely to the fact of his failing memory and physical powers. The problem becomes one of who shall look after him, or of psychiatric treatment if severe. If one follows up the patients who have episodes of anxiety over a period of years, one finds that a number of them become more stable, some continue to have episodes perhaps of greater severity, and a proportion develop the chronic anxiety state. In this the symptoms are the same in principle, but certain of them, psychological or somatic have become habitual. Where it is a somatic reaction that has become habitual, the psychological symptoms are less obvious and exacerbations are not necessarily related to environmental stress. We now have to deal with syndromes like irritable or spastic colon, neurodermatitis, or some forms of chronic dyspepsia. Organic changes may have supervened like hyperthyroidism, duodenal ulcer or ulcerative colitis, although these diseases can be reached by other routes in other patients. The psychiatric picture is no longer one of pure anxiety as hysterical and other mechanisms may have developed, and other personality traits are incorporated in the illness. Most of the treatment has to be along ordinary medical lines, but we can, by explanation of the symptoms, still help the patient to accept the illness for what it is, and prevent it from playing too large a part in his life, and by discussion of difficulties and attitudes help to prevent some of the inevitable relapses. Knowing the normal neurosis of the chronic patient is also helpful in diagnosis as a new symptom is likely to mean organic disease. I recently kept sending such a patient back to hospital on this assumption. She was twice sent back with a diagnosis of "functional" in view of the long history and negative X-ray, but I persisted, and the third time the X-ray showed a carcinoma of the stomach, fortunately found to be still operable. The next group is:

The obsessional personality, which superficially resembles the anxious personality in that they generally come to the doctor for anxiety or depressive symptoms. These are not, however, produced by environmental stress but by a particular attitude on the part of the patient to what may be a normal environment. The patient is dominated by guilt and doubt, so that he is never sure whether he has done right, is constantly checking and rechecking his work, is pedantic, overconscientious and perfectionist. A baby involves a nightmare of minute details which must be exactly right, or promotion at work means intolerable responsibility with the opportunity to make more serious mistakes. This attitude becomes apparent at the slightest discussion, and it is obvious that they are making their own stress. As they are always driving themselves, any physical or environmental change may be enough to precipitate a breakdown into depression, e.g. an influenza, loss of someone on whom they relied, or change of job, especially by promotion or increase in the pace of work. Occasionally they come to the doctor for specific phobias especially fears of certain diseases, or of being contaminated, which may present as a generalized pruritus, or for obsessional doubts, rituals, or compulsions, i.e. what is called the obsessional neurosis when fully developed and which needs psychiatric advice. It should be noted that a recently developed severe obsessional state may be the onset of schizophrenia, psychotic depression or may be due to arteriosclerosis or other brain disease. The handling of the obsessional personality is based on the fact that this type of chronic worrier is incurable by any method open to the G.P., who must be prepared to allow the patient to rely on him for the rest of his life. They can be kept going by confident reassurance constantly repeated with unshakable sympathy and optimism which often works better than explanations. The doctor must try to keep up their confidence by praising their achievements, and be prepared to share responsibility with them and advise them, whether they want to know if the medicine should be taken five or ten minutes after food, or whether Johnny should stay on at school after 15. If they break down with a depression, give them enough time to recover because they always want to go back to work too soon. In this way they can be guided along and remain the useful members of the community which indeed they are.

Lastly I propose to deal with *The hysterical personality*. Hysteria may be defined as the more or less unconscious production, simulation or prolongation of symptoms for an emotional gain. The following is a typical episode: A charwoman of low intelligence and lacking in confidence was told that next day she would have to clean out a disused room. On the way to work next day she collapsed in the bus, was taken home, and sent for her husband. When I saw her she said she had 'flu which had affected her legs so that she could not walk, but there were no physical signs or temperature. As she was no better by the following day, her husband went to her employers and cleaned out the room, whereupon she immediately recovered and said it was due to the marvellous medicine prescribed. Another recent case was a homesick Italian girl who continued vomiting. The absence of other clinical evidence and her attitude to the illness convinced me that she was making her employer play the part of a mother to her through the illness. Her treatment illustrates the treatment of the hysterical episode. One does not make too much fuss of the symptoms or patient

while she is ill, so the girl was told to stay in bed and only drink water, "to give her stomach a rest" and the employer told to stop asking her if she could not eat various delicacies as she had been doing, but to leave her alone with a jug of water. She was also told to mother her as much as possible when she was well to prevent recurrence. At the same time a medicine was prescribed with confident salesmanship and rapid recovery ensued. If there is a temporary problem, it is quickest to let her have what she wants. In the case of the charwoman the husband doing the job of which she was afraid solved it, and the medicine only saved her face, which is equally important.

Anybody can get a hysterical episode, be it only a headache which prevents the fulfilment of an unwanted engagement, and such episodes are normal in childhood. Occasionally they accompany a psychosis or an organic brain disease. The hysterical personality keeps getting such episodes, and the chronic hysteric is never well. If the manifestations are by behaviour, e.g. fits, faints, emotional storms or by dramatic episodes of the type described, diagnosis is easy but pure hysteria is rare in general practice. It is far more common for a patient to elaborate a physical illness for hysterical motives, often with the aid of unguarded remarks by the doctor. There may be a hysterical overlay as in the woman who fails to recover from a confinement and has extraordinarily widespread symptoms from a lumbo-sacral strain or torn and infected cervix, due to the fact that she resents the burden of the baby, wants to punish the husband, or to make sure she does not get another baby, on doctor's advice of course.

There may be a hysterical prolongation of an originally physical illness, e.g. a man who remained unfit for work months after a back injury. I discovered he wanted to break off his engagement, did not know how to tell his fiancée, and was protected from marriage by being unfit for work. After some discussion the engagement was broken off and the back recovered. Another case of hysterical prolongation was a woman who had had an obscure chest illness with a doubtful X-ray shadow. All investigations were negative, including bronchoscopy but a loud and violent cough persisted and when I saw her twelve months later she was still getting certificates. She had lost her husband and money, was untrained for any work, had had a particularly unpleasant job as companion help to an old lady which she had lost as a result of the illness, and was scared of trying again, hence the so-called bronchitis. Sometimes the hysteria is shown only by the attitude to an illness, e.g. the harassed mother who welcomes her influenzal infection, parks the children on her sister, and is convinced she is on the verge of congestion from which she will take some time to recover. Indeed this is what the sympathetic doctor may well have told her and thus given her the right treatment if perhaps for the wrong reason. These cases illustrate the three characteristics of hysteria which must all be present for this diagnosis to be made. These are:

(1) The symptoms must be medically mysterious, i.e. either most of them are not explicable by any pathological condition present or their duration does not follow anticipated lines, i.e. they clear up dramatically with environmental change or suggestion, and they fail to respond to apparently appropriate physical treatments except for temporary suggestion effects.

(2) The attitude to the illness shows a combination of verbal exaggeration with absence of real anxiety. Thus a patient may weep tears of depression and threaten suicide yet laugh heartily the next minute. Pains are described as agonizing, weakness as paralysis, yet the sufferings are accepted with indifference or cheerfulness understandable only if the illness is emotionally satisfying or solves a problem, but has still to be brought loudly to someone's notice.

(3) The illness serves an emotional purpose. This is rarely obvious self-interest, and if we feel contemptuous on diagnosing hysteria we will not get the patient's confidence enough to find out the real motives. Reactions to severe and genuine stress may be hysterical, and a hysteric often cuts off his nose to spite his face.

True conscious malingering is rare, even in the Army in wartime. There is very often an early history of lack of affection or insecurity, and many hysterical illnesses are pathetic bids for attention or protection sometimes carried to extreme lengths, as in the lady who had alleged heart attacks when left, so that her daughter could not go to work or get married, or the one with a "gastric stomach" who vomited if her husband tried to go to his club. Resentment at the husband's behaviour is often the cause, and often justified, but never so much as she maintains. An elder daughter may resent the mother's attention to a younger one, fear of becoming unwanted may precipitate hysteria at the menopause; some women feel compelled to martyr themselves for their families but feel they must show physical stigmata of their martyrdom. There is no symptom which cannot be hysterically simulated, and no shade of emotion or family situation which cannot result in hysterical illness in a predisposed person. A superficially ideal marriage may still be full of frustrations and we should not jump to conclusions without ascertaining the patient's real feelings. A large group of cases depends on outside environmental influences such as work difficulties or compensation in various forms, not necessarily financial compensations.

The elucidation of a case of hysteria may require the most acute clinical acumen and the most subtle psychological insight. In my own practice it has often taken me several months to realize I was dealing with a hysteric, and several years before I understood what the illness was about, and I venture to think we all have some undiagnosed hysterics among our regular clientele masquerading as something else. If we take a long-term view of our patients and periodically review every manifesta-

tion of illness they have had to see what picture emerges, quite often we get a flash of insight and everything clicks into place. If we keep an open mind and do not diagnose hysteria unless all the three characteristics are present, we will not commit the blunder of calling a cerebral tumour hysterical, nor will we make the far commoner mistake of advising operations for minor defects in a vain attempt to cure hysteria. Recognizing the hysterical personality by long-term review is of great value in subsequent diagnosis and treatment, and recognizing its absence is of still more value.

Successful treatment of the prolonged or chronic case is always difficult and sometimes impossible. A life-long hysterical hypochondriac is beyond medical resource. The first essential is not to do harm by unwise suggestion. Hysterics are very suggestible, but they fasten more easily on suggestions that reinforce the illness than otherwise. An operation is the most powerful suggestion that they are physically ill, and such a patient will welcome the idea as it gives her plenty of attention, no responsibility, and makes it certain in the eyes of the relatives as well as herself that she is a severe sufferer. The effect is temporary improvement with subsequent return of all the symptoms, and often some more as well, which she ascribes to the operation itself, and the doctor may put down to adhesions. It is easy to blame minor abnormalities for indefinite symptoms, and E.N.T. and gynæcology offer fertile fields. X-rays may mislead as they may show a gall-bladder which does not fill or an appendix which does not empty, but do not show the personality. A follow-up of 385 cases of appendectomy showed that where there had been no acute attacks, 1% were better and 24% worse. We have all had cases of cholecystectomy without benefit in the absence of biliary colic, and cases of gall-stones that produced practically no symptoms throughout life. One patient of mine saw three eminent gynæcologists in rapid succession. One advised hysterectomy, one induction of menopause by irradiation, and one doing nothing. The difference was in the psychiatric insight of the surgeons concerned, and our problem is often to choose consultants for their psychiatric knowledge rather than for their surgical skill. I have mentally classified my E.N.T. and gynæcological colleagues into the conservatives and those who always operate. I send my hysterics to the former as too often dilatation and curettage, removal of small cysts or fibroids have been followed by my trying to treat interminable gynæcological symptoms like pelvic pains or pruritus. The old term of chronic abdomen for those who have successive abdominal operations has been rendered out of date by the advances in surgical technique. One patient who had already had her gall-bladder, appendix and uterus removed without benefit (and three abdominal scars is diagnostic of hysteria) had her heart opened, and closed again when valvulotomy was found not to be required; another had an unnecessary ventriculography. In these cases the fault was mine for not being sufficiently emphatic about the patient's personality in my letter to the hospital, as surgeons cannot be expected to understand our patients in the way we do after years of observation.

Elaborate investigations can reinforce the illness. If the patient is known not to be of hysterical personality, one cannot be too careful in investigating a new illness, but in the case of a known hysteric, we have to judge between aggravating a neurosis and the fear of missing something. If what is feared is major, e.g. a carcinoma, it must be excluded, but if what we are looking for is of a minor kind it is better to miss it.

Investigations of indefinite symptoms often give indefinite results and make the patients believe they have an obscure illness that puzzles the doctors. A limited investigation, e.g. X-ray for ulcer, may be unavoidable, but, once done, the case should be handled along other lines and not discussed with the patient in terms of high or low stomach or other items of the radiologist's report. Giving symptoms a physical label like this is readily seized on by the patient. Similarly patients who are treated for chronic fibrositis at rheumatism clinics for months without permanent benefit believe they have a disease of the muscles so bad that even electricity cannot help them, and the same applies to manipulation by orthopædic surgeons. The principle is that once we have diagnosed a hysterical personality, we must fight to prevent them from having physical treatment of a kind which will reinforce the illness by suggestion.

The hysteric will resent being told there is nothing the matter, and will not accept theories of psychological causation without training. If we play down the physical side and say it is aggravated by nervous tension, suitable ones may be encouraged to talk of their resentments or frustrations. Over a series of interviews they often drop the physical illness themselves or are satisfied with very little physical treatment if we show we regard the other side as more important, and are then quite happy to give us a somewhat one-sided account of their troubles. Where there is real environmental stress, help along the lines discussed under anxious personality will be useful, but more often we have to deal with personality difficulties leading to abnormal reactions in a more or less normal environment. In such a case we must generally be satisfied to do as little harm as possible, and if we can prevent a part-time hysteric from becoming a full-time one we have done a good deal. Deep psychotherapy and attempts to alter fundamental attitudes are outside the scope of general practice. They are immensely time-consuming as such a patient will not accept the most obviously rational and common-sense advice unless preceded by long and intimate talks. This carries the risk of the doctor getting incorporated into the emotional situation with such complications as improvements and relapses, depending on whether the patient is falling in love or out of love with the doctor. Without attempting formal psychotherapy, the doctor who understands his patient's reactions can

encourage healthier attitudes and interests, and over a long period do quite a lot of good. Some can be sent to a psychiatric clinic, and although only a proportion will be accepted as suitable for prolonged treatment, the psychiatrist's report on the nature of the patient's problems will be helpful to us. It is no good trying to get them all treated at the clinic, we will only be told there is a long waiting list for treatment and the patient is too chronic to benefit anyway. It is often more helpful to try and treat the relatives. One difficulty is that there is frequently a family neurosis which encourages the growth of hysteria. The wife who wants to dominate seems to have married a man who takes pleasure in being dominated by a sick wife, if she wants to be a frail martyr to her health her husband or daughter are over-conscientious in their ministrations. Nevertheless it is often possible to educate the relations in the principle of giving her affection and attention when she is well, practically ignoring her when she is ill, and not allowing her illness to interfere with their legitimate activities. A daughter must not be allowed to sacrifice her life to a demanding mother. A psychiatric colleague of mine cured a chronic hysteric without seeing her entirely by treating the neurosis of the over-conscientious daughter, so that the mother was forced to find healthier outlets.

Some symptoms like hysterical faints or fatigue can be ignored, but troublesome ones like vomiting or pruritus need some form of suggestion whether long-term treatment be undertaken or not. Unfortunately patients nowadays are mostly too sophisticated to believe in the value of strong smelling medicines like valerian, but these can still be used for the more simple-minded ones, especially if given with a little sales talk like "This will cure if only you can take it, but it's got a very strong smell, you had better go into the kitchen when you take it". Highly coloured paints are useful, and one dermatologist told me he got his best results in pruritus vulvæ by giving X-rays without turning on the machine. Suggestive methods are most effective if the doctor believes in their value, so new remedies introduced with a fanfare of advertising matter will have temporary success, and quacks will cure some of our failures. Whatever method is used, unless some alteration in the patient's attitudes or environment is possible, the condition will generally reappear, perhaps in another form.

It must be pointed out that only medical aspects of personality have been discussed. To say someone is of anxious or hysterical personality is like saying he is a duodenal ulcer or chronic bronchitis. Other aspects may be more important, e.g. Charles Darwin might nowadays be called a gross hysteric but he was a creative scientist as well, and many talented artists, especially in the theatrical profession, have more than a touch of hysteria. Some obsessionals are the backbone of industry and the Civil Service would not work without them.

Conclusion

The G.P. is often exhorted to study the whole man. What I have tried to emphasize is that the whole man is the patient not only in his setting but in time. Taking the long-term view need not be impossibly time consuming. Patients who have found that when they need help the doctor is prepared to give them ten or fifteen minutes will be satisfied with two minutes on another occasion when the need is less. With experience we pick out the patients we can help and cut down the time spent on those we cannot, so that the overall average remains reasonable—in my own case four minutes per patient in the surgery. By long-term reviews of the patient, observing similarities and recurrences in his illnesses and thus identifying personality, we not only become more efficient doctors but we add new interest to our work.

Dr. M. Narasimha Pai, Consultant Psychiatrist, Belmont Hospital, Surrey:

At the Belmont Hospital (Neurosis Centre), where every year about 1,500 patients are admitted on the recommendation of consultants, the psychiatrists have opportunities of checking the patient's physical and mental state on admission, and of following-up the patient over a number of years after discharge from hospital.

Generally speaking, no psychiatrist to-day can claim to make an accurate diagnosis without (*a*) excluding organic diseases, and (*b*) without possessing the following information: (i) an independent social history incorporating details about the patient's family, the developmental history, childhood neurotic traits, and school and work record, (ii) efficiency and personality reports from employers, (iii) details of the onset of the illness and of any obvious environmental or other stresses, and (iv) assessment of the patient's mental state as a result of one or more interviews. Sometimes the following additional information is also essential: (v) results of intelligence and psychological tests, (vi) behaviour of the patient while under observation, (vii) results of biochemical and electro-encephalographic investigations in appropriate cases, (viii) reactions to test situations, and (ix) reactions noted when drugs such as barbiturates, methedrine, &c., are given intravenously. Information obtained from these and several other sources (Pai, 1946), which may be compared to the pieces of a jig-saw puzzle, is integrated and the picture which emerges forms the basis of a psychiatric diagnosis. The diagnosis is not based on mere symptomatology but on psychological factors, mental mechanisms and motives. To discover environmental stresses the help of specially trained psychiatric social workers is enlisted.

There is now an increasing tendency on the part of general practitioners to call patients neurotic and to apply psychiatric labels without making as sure as possible that the symptoms do not arise

from some organic cause. By a psychoneurotic or functional illness we mean the disturbances of function of an organ or system in the absence of physical changes to account for them. The ability to exclude organic diseases depends partly on one's clinical experience and partly on the methods of investigation employed. It is therefore obvious that a practitioner without psychiatric training who undertakes this special work shoulders a great responsibility beset with dangers, and mistakes in diagnosis are inevitable. First, when unemployment, financial stress, marital discord or housing problems are obvious the practitioner may dispense with a physical examination and may wrongly call the patient neurotic. Secondly, a psychiatrist may make a wrong diagnosis and communicate this to the practitioner who then undertakes treatment on his own initiative with perhaps serious consequences. Thirdly, while carrying out the treatment indicated by a psychiatrist new symptoms may arise which the practitioner ignores on the supposition that they, too, are psychogenic.

Numerous instances could be given of patients suffering from advanced cancer (Pai, 1950*c*, 1954; Richardson and Russell, 1952) and other organic diseases (Pai, 1949, 1950*a*, 1950*b*) who had been wrongly given psychoneurotic labels and treated as such—first by the general practitioners and then by the consultant psychiatrists at the London hospitals. Yet any old-fashioned general practitioner, uninfluenced by modern psychiatric literature, might easily have guessed the organic nature of their illness by routine examination.

Practitioners are also apt to underestimate the intelligence of their patients and to forget that the vast majority of the population already know something about the symptomatology of the nervous disorders and their treatment. The daily papers, the psycho-analytic literature in public libraries, the radio, the cinema and the television cater for them. It is no wonder that the public has become psychology-conscious and that the physically ill patient postpones going to his doctor until the symptoms have disabled him for work and he may then mislead the doctor by giving a long list of obviously neurotic symptoms. The genuine neurotic may also delay consulting his doctor until his symptoms have consolidated or he may withhold useful information. He may then need something more than reassurance and explanation.

If some practitioners claim to give psychiatric treatment, e.g. analysis, there may be a demand on the part of patients of other practitioners for similar treatment whether indicated or not; this is happening in the case of hypnosis. All over the country some patients are pestering their practitioners for hypnotic treatment and those doctors who cannot offer this run the risk of losing them.

Modern treatment of many of the psychiatric disorders involves not only the treatment of the patient but also the control of the environment and modification of the socio-economic factors—activities for which the general practitioners possess neither the time nor the facilities. Even the administration of barbiturates, endocrine preparations and vitamins is not without risk, as complications may arise, causing distress to the patients and their relatives. In some of the males referred to me it was found that well-marked gynæcomastia, uncontrollable homosexual tendencies and behaviour disorders had resulted from stilbœstrol prescribed by the practitioners.

Compensation and pension cases present special difficulties. As patients are fully aware that compensation depends on the length of period they remain on the sick list, no matter what treatment is given, they may persist in complaining of symptoms until their claims are settled. Disability and war pensions are likely to be discontinued or reduced, if pensioners do not complain of symptoms, or if they do not attend for treatment. Soldiers granted pensions for psychoneurosis during the First World War are still attending for treatment; the frequent attendance of such patients in the surgery and the consequent gossip may lower the morale of other patients and deter at least some of them from seeking the practitioner's advice.

There are also the legal aspects. If a practitioner, as a result of errors either in diagnosis or in treatment, lays himself open to legal proceedings for negligence, then the sympathies of the judge and jury are likely to be with the patient and his relatives, and heavy damages against the practitioner must be expected. Such successful legal actions may well encourage the chronic neurotics to bring vexatious proceedings against their psychiatrists and thus hinder further progress in psychiatry.

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Clinical Section

President—JAMES L. LIVINGSTONE, M.D., F.R.C.P.

[*March* 12, 1954]

Sarcoidosis, Hypercalcæmia, Calcinosis and Renal Impairment.—K. M. CITRON, M.D., M.R.C.P. (for J. G. SCADDING, M.D., F.R.C.P.).

Miss I. E., aged 23.

In 1948 she commenced work in a factory, and was engaged for one year in coating the inside of fluorescent tubes with a mixture containing beryllium phosphor and nitro-cellulose. At the end of this time an X-ray of her chest showed no abnormality. Ten months later she complained of dry cough and dyspnœa on exertion. On examination, there were no abnormal physical signs. However, the chest X-ray showed miliary mottling throughout both lungs and clouding at the right hilum (Fig. 1). The Mantoux was negative 1 : 100 O.T. and the E.S.R. was 75 mm. in one hour (Westergren). The following month she developed erythema nodosum on both legs and small raised nodules on the skin of the hands at the site of old scars. A biopsy of one of these lesions showed that the corium was involved by accumulations of epithelioid cells, among which were multinucleate giant cells. There was no caseation and the appearances were those of sarcoidosis. Beryllium was not demonstrated in the skin section by means of histochemical staining techniques.

In 1952 a painless bulbous swelling developed on the right fourth toe. X-ray of this showed a calcified excrescence on the terminal phalanx (Fig. 2). As the swelling was interfering with her walking, the terminal phalanx was amputated. The section showed a granulomatous reaction around deposits of calcium.

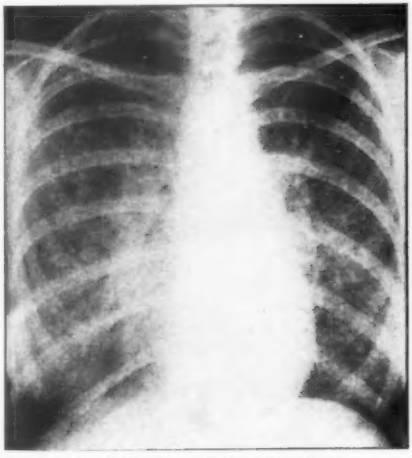

FIG. 1.—Miliary mottling of the lungs.

FIG. 2.—Calcified excrescence on the terminal phalanx.

In July 1953 she complained of some itching of the eyes and also slight cough and dyspnœa and frequency of micturition.

On examination.—There were many very small white raised nodules in the conjunctivæ. These ocular appearances were first described by Walsh and Howard (1947) in association with hypercalcæmic states and were attributed to deposits of calcium phosphate in the conjunctiva. They noted that the lesions might disappear after the correction of the hypercalcæmia. More recently the same condition has been described as occurring in sarcoidosis with hypercalcæmia.

The tympanic membranes showed extensive white calcareous areas. The skin was normal and there were no palpable lymph nodes. The liver was palpable. Blood pressure was normal.

Investigations.—Chest X-ray showed no change and tomography confirmed hilar lymph-node enlargement. No tubercle bacilli were found in gastric lavage. *X-ray hands*—1952: Showed trans-

lucencies in the tips of the terminal phalanges and several dense opaque deposits in the soft tissues of the fingers. 1953: The X-ray was similar except that the deposits in the soft tissues were absent. *X-ray abdomen* showed two calcified areas in the left kidney. The urine contained a trace of albumin and hyaline, cellular and granular casts. Culture was sterile. Fig. 3 shows the biochemical findings. The serum calcium was remarkably high (19 mg./100 ml.) and the inorganic phosphorus was also raised. The blood urea was high (50 mg./100 ml.) and the urea clearance was reduced to 55% of normal.

Treatment.—She was given isoniazid and streptomycin. The dose of the latter was reduced to 0·5 gramme on account of high blood streptomycin level due to renal impairment. No significant change in the biochemistry occurred during one month of this treatment. Cortisone was then given 150 mg. a day by mouth for ten days and thereafter 100 mg. daily. The diagram shows the rapid fall in serum calcium and phosphorus which took place. The urinary calcium excretion also declined. She is now maintained on a dose of 25 mg. of cortisone daily and three months after the commencement of treatment with this hormone her calcium is between 12 and 13 mg./100 ml. and renal function has shown marked improvement. The blood urea is 25 mg./100 ml. and the urea clearance is now 96% of normal.

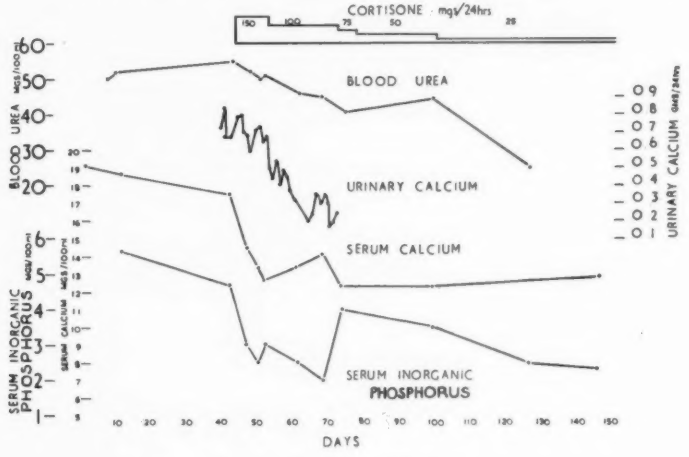

FIG. 3.—Biochemical findings.

There has been improvement in the appearance of the eyes. There is no frequency of micturition. Though there has been no appreciable change in the radiological appearance of the lungs, dyspnœa on exertion is less.

Comment.—Since beryllium was not demonstrated in the skin lesion, and erythema nodosum is not recorded as a feature of beryllium sarcoidosis, the relationship of her disease to exposure to beryllium phosphor is not established.

Whilst hypercalcæmia in sarcoidosis is not uncommon, renal impairment is rare. In those cases in which calcium metabolism has been investigated, hypercalcæmia and hypercalciuria have been demonstrated, and renal impairment is thought to be due to nephrocalcinosis (Klatskin and Gordon, 1953). The response of the hypercalcæmia to cortisone was prompt, and was associated with improvement in renal function. Similar findings in one case have been reported by Dent *et al.* (1953). Since hypercalcæmia in sarcoidosis may lead to renal impairment, and can be controlled with cortisone, its recognition is of practical importance.

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Dr. C. E. Dent: We are following at University College Hospital 4 cases of sarcoidosis complicated by hypercalcæmia. They are now all under continuous cortisone treatment, but as adequate supplies have only been available since November 1953, some cases were, at first, treated intermittently. All the cases had symptoms of constitutional disturbance, mainly tiredness, anorexia and lack of energy, but 2 had severe pain, 1 vaguely localized in the epigastrium, the other in the

lower back and thighs, and 1 had considerable nausea and tendency to vomit. They all had varying degrees of renal failure with blood urea in the range of 40–150 mg./100 ml. We attribute this to the toxic action of the raised calcium level. We found that the hypercalcæmia always returned to normal after a short course of cortisone. We now begin with 150 mg. a day for ten days, gradually lowering this later, using as the desirable criterion a plasma calcium of less than 11·0 mg./100 ml. Coincident with this fall of the plasma calcium level the patients' symptoms have improved in the most spectacular manner, there has been some improvement in renal function and all have been able to return to normal activity (window cleaner, schoolmaster, apprentice, housewife). Finally in those earlier cases in which the cortisone had been stopped the plasma calcium soon rose to abnormal levels with the return of symptoms and a worsening of renal function.

The maintenance dose of cortisone has been between 25 and 75 mg. a day. The dosage appears to be fairly critical as one patient deteriorated clinically and biochemically on 25 mg. a day and another on 37·5 mg. Both, however, seem now to be satisfactorily maintained on 37·5 and 75 mg. respectively.

We are naturally interested in the apparently irrelevant coincidence of a definite biochemical defect in occasional cases of this disease of obscure ætiology. The hypercalcæmia does not appear to be associated with any particular clinical manifestation of sarcoidosis. Our 4 cases are quite unlike each other in the localizations of the readily detectable sarcoid deposits. The hypercalcæmia, however, with its associated symptoms and renal dysfunction, must be related to the other manifestations (skin, glands, chest, eyes, &c.) which occurred, since they all appeared to be controlled adequately by the same dose of cortisone.

We are studying the pathogenesis of the hypercalcæmia. In a preliminary publication (Dent, *et al.*, 1953) it was suggested that active bone dissolution due to small sarcoid deposits was responsible, as occurs with multiple osteolytic tumours. This would explain well its ready reversibility with cortisone and its approximate coincidence in time with the regression elsewhere of the sarcoid lesions. It also explains well the symptoms in one patient which closely resemble those of bone pains from the lower lumbar vertebræ. However, a further disorder of calcium metabolism is now coming to light as we begin to apply calcium balance studies to the problem (Anderson *et al.*, 1954). This is the unusually high absorption of calcium from the gut which seems to be a constant finding in these cases. Naturally this, too, will flood the blood stream with calcium and will raise the blood level in the absence of adequate compensatory mechanisms. This high absorption is impossible to explain on the basis of our present knowledge of calcium metabolism, and it seems remarkable that it should occur as a complication of a granulomatous condition. The action of cortisone in apparently reversing the metabolic defect is also as obscure as it is fascinating, but we appear to have enough evidence already to acknowledge a still further pharmacological action of this versatile hormone. This action may not, of course, be due to a direct effect on calcium metabolism but may be indirect as the result of the action on the sarcoid granulomas.

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Idiopathic Osteoporosis—Symptomatic Relief by Plasma Infusions.—J. ANDERSON, M.B. (for C. E. DENT, M.D.).

Mr. W. A., aged 30.

November 1947: Sharp pain in right hip on exertion. X-ray showed no abnormality.

March 1948: Severe lower back pain on exertion. Treated with physiotherapy.

December 1948: Felt a lump on his spine and was admitted to the West Middlesex Hospital under Mr. E. A. Devenish. In view of the radiological and other findings he was referred to University College Hospital for further investigation.

January 1949: X-ray: gross rarefaction, loss of trabeculæ and wedging of lower thoracic and upper lumbar vertebræ. Plasma calcium 10·3 mg./100 ml. Plasma inorganic phosphorus 4·1 mg./100 ml. Alkaline phosphatase 9·3 K.A. units. Blood biochemistry and renal function normal. Fitted with a spinal support and treated with calciferol and calcium lactate. Until 1951 he had repeated attacks of acute pain in the back and ribs. His general health remained excellent.

September 1951: Sudden præcordial pain radiating to both axillæ. Readmitted to hospital. He had lost about 7 in. in trunk height during the past few years. Blood biochemistry, 17-keto- and 11-oxy-steroid excretion normal. His back pain had been so severe while at work (carpentry) that he was forced to turn to lighter work and the hospital almoner therefore arranged for him to be trained as a glassblower. Treatment with methyl testosterone and then with stilbœstrol did not bring about symptomatic relief or alter his continuing negative calcium balance of 200 mg./24 hours.

He was then given 32 bottles of plasma intravenously over a period of three weeks. This resulted in a decrease of his urinary calcium excretion from 200 mg. to 20–50 mg./24 hours (Fig. 1). He had a febrile reaction to the first plasma infusions, but this was controlled in subsequent infusions by large doses of aspirin.

Symptomatic improvement began in about two to three weeks and continued for some months. After this the pains became worse and his urinary calcium slowly rose to 100 mg./24 hrs. He has been readmitted on three occasions since then on account of worsening symptoms, and on each

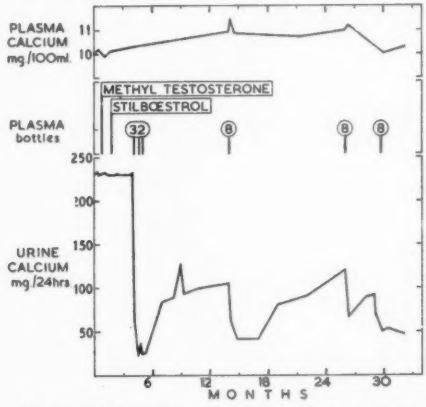

FIG. 1.—Diagram showing fall in urine calcium independent of plasma calcium level on giving intravenous plasma (case W. A.).

occasion he has been given intravenous plasma and his severe symptoms have been relieved for a period of months. Improvement has coincided each time with a decreased urinary calcium excretion. His biochemical investigations have remained otherwise normal.

Albright and Reifenstein (1948) have found similar changes in the treatment of idiopathic osteoporosis with intravenous plasma albumin.

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Lingual Thyroid.—R. ALHADEFF, F.R.C.S. (for M. R. EWING, F.R.C.S.).

Mrs. K. L., aged 32. Secretary.

History.—In August 1953, after a tooth extraction, she developed an infected socket for which pencillin lozenges were prescribed. While taking the latter she developed a sore throat and first became aware of a swelling on the back of her tongue. It was quite painless, only very rarely did it cause her a choking feeling during the day and it never distressed her at night. It did not become larger in the ensuing months although she noticed a slight variation in size from day to day. It was unaffected by her periods which were regular. There was no difficulty in swallowing, breathing, or speech, and the tumour did not bleed. She was first seen at Hammersmith Hospital on 16.1.54, complaining only of a swelling on the back of the tongue. She was in all other respects fit and well.

On examination.—Her tongue presented a tumour 2 cm. in diameter, occupying the posterior third and projecting into the oro-pharyngeal isthmus, to impinge against the uvula (Fig. 1). The tumour was pinkish in colour; its surface was smooth and covered by intact vascular mucosa. On palpation it was fleshy and extended deeply into the substance of the posterior third of the tongue. In the neck neither the isthmus nor the lobes of the thyroid could be felt. Clinical assessment indicated that the patient was euthyroidic.

Investigations.—A soft tissue film of the neck (Fig. 2) revealed a shadow at the base of the tongue bulging into the pharynx and displacing the epiglottis slightly backwards and downwards.

A tracer dose of 100 μc. I^{131} was given by mouth and a urinary excretion test together with a neck scan were performed under the care of Mr. Selwyn Taylor. At the end of a forty-eight hours period 51·3% of the total intake of radioiodine was excreted in the urine. This figure showed that thyroid function was within normal limits.

Localization studies were carried out by means of a collimated Geiger counter. Maximal activity was recorded when the counter was directed towards the upper end of the thyroglossal track. Little activity was obtained through the open mouth, as it was a difficult problem technically to collect parallel rays in this location. No activity was detected below the level of the hyoid bone (Fig. 3).

On the basis of these findings a diagnosis of lingual thyroid was made.

Comment.—Fig. 4 illustrates the various positions that may be assumed by thyroid tissue in the line of the thyroglossal duct, a derivative of the first pouch, originating just caudal to the tuberculum impar.

Lingual thyroid is a rare anomaly. Verneuil first described this condition in 1853 and by 1936

Montgomery collected from the literature and reviewed 144 authenticated cases. How much the ultimobranchial bodies from the fourth pouch contribute to the lateral lobes of the gland is debatable, although Weller (1932) maintains that they take a considerable part in their formation. In our patient there is no active thyroid tissue in the neck derived from the fourth pouch.

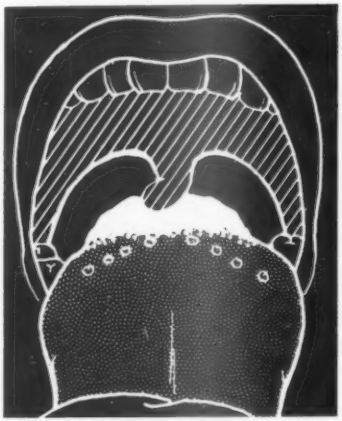

FIG. 1.—Lingual thyroid projecting into pharynx from posterior third of tongue.

FIG. 2.—Soft-tissue film showing tumour displacing the epiglottis slightly backwards and downwards. (Epiglottis and hyoid bone are partly superimposed.)

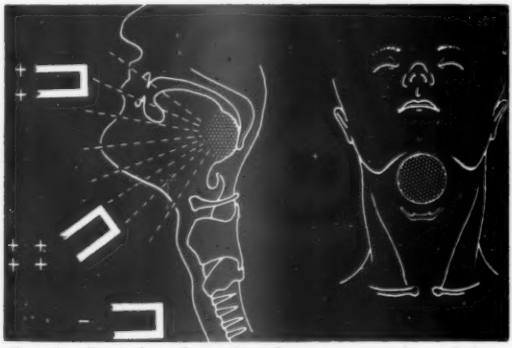

FIG. 3.—Intensity of uptake of gamma rays by a Geiger counter at different levels.

FIG. 4.—The thyroglossal duct and common positions of ectopic thyroid tissue.

Treatment.—It is generally agreed that surgical removal, whether partial or complete, is not justified in the absence of symptoms. If, on the other hand, the thyroid could be prevented from enlarging or even made to regress by medical measures alone, that would be the treatment of choice.

The patient is therefore being given l-thyroxine sodium, 0·1 mg. twice daily, in order to diminish the body's demands on the thyroid, and iodized salt prophylactically, to counteract the goitrogenic effects of a possible iodine deficiency in her diet.

It is hoped by these measures to keep the size of the gland under control.

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Mr. Selwyn Taylor: The radioactive iodine investigations on this patient left no doubt that all the thyroid tissue was situated at the base of the tongue, but since the surface presenting into the mouth was small in

extent and showed no likelihood of ulceration or hæmorrl age, it was thought reasonable to prevent the thyroid becoming larger rather than to excise the tissue. For this reason the patient is being given l-thyroxine by mouth and she herself already has volunteered the information that the swelling is smaller.

Coarctation of the Aorta with Calcified Aortic Aneurysm.—BETTY WALKER, M.A., M.B., M.R.C.P. (for BERTRAM JONES, M.D., M.R.C.P.).

L. M., aged 47. Housewife and skirt finisher. 2 children aged 18 and 16 years.

History.—Admitted to hospital aged 9 years with rheumatic fever. High blood pressure (145/100–170/100) was noticed during her first pregnancy. Short rest periods in hospital were followed by normal confinements at term. Since that time, she has continued symptom free. In addition to household duties, she undertakes light work for four to six hours a day. In December 1953 she attended hospital on account of a minor ailment.

Examination.—Medium build and height. C.V.S.—The radial pulse is regular. B.P. right arm 230/110; left arm 225/110. There is marked pulsation of the arteries of the neck. Pulsation is prominent in the episternal notch where a large vessel can be felt. Pulsating tortuous vessels about the size of normal radial arteries are present in both interscapular regions. Anastomoses can be traced into the right suprascapular fossa. Pulsating vessels are present also in both infrascapular fossæ and at the scapular angles. Soft murmurs in late systole can be heard over these vessels. Similar murmurs are heard on either side of the sternum continuing over the upper part of the abdominal wall. There is an apical and basal systolic murmur. The femoral pulses, although delayed, are easily palpable as are those of the tibial and dorsalis pedis arteries.

Investigations.—Teleradiography and cardioscopy: Marked notching of the inferior borders of the ribs is present. There is moderate enlargement of the left ventricle with prominence of the ascending aorta to the right in the antero-posterior view. A calcified aneurysm replacing the normal aortic shadow lies below the origin of an enlarged left subclavian artery (Fig. 1). Electrocardiography shows left axis deviation with left ventricular preponderance. Urine normal. Blood urea 37 mg./100 ml. W.R. negative.

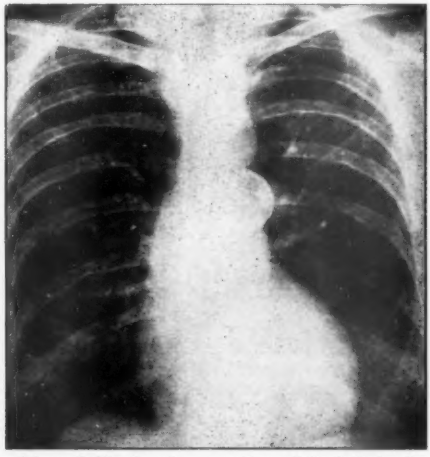

FIG. 1.—P.A. view of heart and lung fields showing notching of the inferior borders of the ribs and calcified aortic aneurysm.

Comment.—The points of interest in this case are (1) The well-being of the patient and the absence of symptoms at a late age. It is instructive to recall that she has withstood what appears to have been a genuine attack of rheumatic fever in childhood and the physical stress of two pregnancies and confinements, at the latter end of the third and the beginning of the fourth decade, without sequelæ.

(2) The extreme degree of coarctation demonstrated by a remarkably extensive collateral circulation which has succeeded in establishing a normal blood supply to the lower limbs.

(3) The association of the coarctation with an unusual calcified aneurysm distal to the coarctation. The radiological appearances in the different views suggest that the deposition of the calcium has resulted in the formation of an almost symmetrical self-limiting capsule.

Mixed Parotid Tumour.—IRENE S. CADE, F.R.C.S. (for GEORGE QVIST, F.R.C.S.).

Mr. W. J. G., aged 72, referred by Dr. D. M. Cockell, has a parotid tumour which has increased in size gradually over a period of eighteen years. The tumour is large, mobile, lobulated and encapsuled with no evidence of infiltration of skin, lymph glands or facial nerve (Figs. 1 and 2).

FIGS. 1 and 2.—Mixed parotid tumour—eighteen years' history.

The average length of history prior to treatment in most series is seven to eight years, and the majority of tumours reach a size of from 5 to 10 cm. in diameter. Occasional cases are reported in which the tumour has been present for twenty to thirty years and has reached a large size.

Ahlbom, in 1935, reported a patient in whom a tumour weighing 4½ lb. was removed after seventeen years.

FIG. 3.—The tumour excised.

The incidence of malignant change in mixed parotid tumours is disputed. According to Patey (1940) there is no proof that alteration in type from a mixed parotid tumour to a squamous-celled or glandular carcinoma ever takes place. Malignant characters may, however, be assumed by previously benign tumours without change of histological type. This is not uncommonly found in recurrences following surgical excision, but is very rare spontaneously.

The present case illustrates the prolonged course of untreated benign parotid tumours without the development of malignant change.

Addendum.—The tumour was removed on 8.4.54, in collaboration with Mr. C. E. Shattock. It shelled out easily, there was little bleeding and the facial nerve was not seen but could be felt deeply against the neck of the mandible. The tumour weighed 4 lb. 6 oz. and measured 19 × 15 × 13 cm. (Fig. 3). Section showed no malignancy. Post-operative course uneventful.

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Pancreatic Lithiasis. Steatorrhœa. Diabetes Mellitus. Pulmonary Tuberculosis. Intermittent Claudication.—M. MENCER MARTIN, M.D., M.R.C.P. (for J. L. LIVINGSTONE, M.D., F.R.C.P.).

T. W., male, aged 52, admitted early in January 1954 for investigation of diarrhœa and abdominal discomfort of three years' duration.

History.—Sudden onset of diarrhœa in January 1951 with 4–5 motions per day and 1 or 2 at night. Easily tired and exhausted. The stools were fluid to semi-formed, bulky and tended to be pale and float on water. Never any mucus or blood. Three months after the onset of the diarrhœa he developed right-sided pleurisy and was discovered to have pulmonary tuberculosis with infiltration of the right upper lobe. He was treated with bed-rest, streptomycin and PAS and in October 1951 he was admitted to Lenham Sanatorium. Sputum became negative and in November 1951 a right A.P.

was induced. He was discharged from the sanatorium in December 1952, since when he has attended the Camberwell Chest Clinic regularly for refills. The treatment for his pulmonary tuberculosis had no effect upon the diarrhœa. At one stage it was thought to be due to PAS, but there was no remission after the drug was stopped. In the summer of 1953 intermittent claudication in the right calf forced him to stop walking after about 100 yards.

Although the tuberculous disease responded satisfactorily to treatment, the patient failed to regain the 2 st. in weight lost earlier on.

Past history.—None of abdominal pain or thirst or polyuria.

Examination.—There were no abnormal physical signs apart from absent pulsations in the femoral, popliteal, dorsalis pedis and posterior tibial artery on the right side. Pulsations were easily palpable in the abdominal aorta and in the arteries of the left leg. B.P. 160/90.

Investigations.—Urine: occasional traces of sugar. Stool: Microscopy: A few red cells, partially digested muscle fibres and moderate number of fat globules. Culture negative including *M. tuberculosis*. Blood: Hb 106% (15·7 g.%), W.B.C. 11,000, 59% neutrophils, 3% eosinophils. P.C.V. 49%. E.S.R. 9 mm. in the first hour (Wintrobe). Plain X-ray abdomen: Calcification present in the region of the pancreas (Figs. 1 and 2).

FIG. 1. — Plain X-ray abdomen showing pancreatic calcification.

FIG. 2.—Lateral X-ray of abdomen showing pancreatic calcification.

FIG. 3. — Barium meal X-ray showing doubtful coarsening of normal feathery appearance of small intestinal pattern only.

Biochemistry: Blood urea 31 mg./100 ml. Total serum proteins 6·45, serum albumin 4·24, serum globulin 2·21 grammes%; A/G ratio 1·91:1; serum calcium 9·4 mg./100 ml. Plasma phosphorus 3·4 mg./100 ml.

Fat balance showed 72% and 59·6% absorption on a 50 grammes/day intake estimated over four days each. Average percentage absorption therefore over eight days 65·8%.

Duodenal juices: Diastatic activity was 8,760 units/ml., but there was no significant lipase or tryptic activity. No diastatic activity in urine or blood. Tryptic activity in stool present only in a dilution of 1 in 4, i.e. trypsin absent.

Glucose tolerance curve: Fasting blood sugar 124 mg.%. Half hourly blood sugars after 50 grammes glucose by mouth were 185 mg.%, 270 mg.%, 262 mg.% and at two hours 262 mg.%.

Barium meal X-ray: Calcification present in the abdomen, pancreatic in site. Small intestinal pattern showed some doubtful coarsening of the normal feathery appearance unlike that seen in idiopathic steatorrhœa. There was some tendency to flocculation and segmentation (Fig. 3).

Cholecystogram was normal.

X-ray pelvis, thighs and legs showed no arterial calcification.

Oscillometry:

	R.	L.
Thigh	½	5
Calf	½	3½
Ankle	>¼	1

Reflex vasodilatation test: Normal response in both great toes, the temperature rising rapidly from 20° C. to above 33° C. within 40 minutes. The calves behaved alike and cooled from about 32° C. to 29° C. The right thigh was 2° C. below the left thigh at the start of the experiment and although its temperature rose by about 1° C. it remained nevertheless 2° C. lower than the opposite thigh.

X-ray chest showed a right A.P. with a thickened pleura. On screening there was little movement.

Conclusions.—The laboratory findings suggested both acinar deficiency and insular deficiency and in conjunction with the radiological appearances confirmed the diagnosis of pancreatic lithiasis with impairment of pancreatic function.

The intermittent claudication is believed to be due to a block of either the right internal or common iliac artery which appears to be confirmed by the subsequent progress.

The presence of pulmonary tuberculosis is thought to be unrelated to the pancreatic deficiency.

Progress.—The patient was put on a diet containing 70 grammes of fat, 200 grammes of carbohydrate, and 100 grammes of protein daily and was given Pancreatin forte 120 grains daily. On this regime his urine remained sugar free, he lost all abdominal discomfort, claimed to feel in perfect health and had, on the average, two bowel motions per day, with soft, but formed stools which he considers his normal habit. Whilst on treatment estimation of fæcal fat showed 95% to be split.

His exercise tolerance increased whilst under observation and he became able to walk 300 yards at a brisk pace in three minutes before the onset of claudication and even then he was able to continue walking.

The right A.P. was refilled at fortnightly intervals. The final decision with regard to this "frozen" A.P. will have to be taken in the future.

Comment.—Pancreatic calcification was described by De Graaf as early as 1667 but it was only in 1903 that Allen published the first complete clinical and pathological study of diffuse pancreatic lithiasis. While the disease was once considered a relatively rare condition, the diagnosis is being made with increasing frequency owing largely to the more frequent use of radiography and the greater awareness of the condition by clinicians. Thus Fisher (1946) collected 200 cases described up to 1946 and since then the number has increased to well over 300. Nevertheless pancreatic calcification is not commonly seen and the aggregate figures from 10 pathological departments showed only 31 cases among over 117,000 autopsies.

The interest in the present case lies in the fact that no history suggestive of previous attacks of pancreatitis which is generally believed to be the cause of pancreatic calcification can be obtained. Although it is possible that there could have been attacks of abdominal pain in childhood, which had been forgotten, the time interval would appear to be too great, the longest period on record between abdominal pain and the appearance of pancreatic calcification being twenty-two years (Gambill, 1949). In Gambill and Pugh's (1948) series of 39 cases from the Mayo Clinic, two-fifths with pancreatic calcification had no history of previous pancreatitis and no serious impairment of pancreatic function. Gambill and Pugh found a high correlation between a previous history of pancreatitis, extent of pancreatic calcification and the incidence of diabetes mellitus and steatorrhœa.

In previous publications a difference was made between "diffuse calcification", "disseminated calcification", "mixed calcification" and "multiple calculi". However as Peters *et al.* (1951) have suggested no distinction should be made since the clinical course and, in particular, the pathology are similar. Histologically all cases reported appear to be cases of pancreatic lithiasis with findings characterized by acute and chronic inflammatory changes, diffuse replacement fibrosis, pseudocysts, dilatation of ducts and intraductal calculi. Calcification, according to Edmonson *et al.* (1950), is due to precipitation of calcium carbonate from supersaturated pancreatic juice.

Diabetes mellitus is the commonest complication of pancreatic lithiasis occurring in up to 75% of cases. Steatorrhœa appears to be less common. Pulmonary tuberculosis has been reported in 10 to 15% of cases in various series in the literature, yet there is no evidence that the two are in any way related, unless the presence of diabetes mellitus or malnutrition predisposes to tuberculosis.

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Section of Proctology

President—T. McW. Millar, F.R.C.S.Ed.

[*January* 13, 1954]

The following cases and specimens were shown:

Perineal Heterotopia of Rectal Mucosa.—Mr. Peter W. Ingram.

Demonstration of Paintings of Sigmoidoscopic Appearances.—Dr. N. F. Coghill.

Spleen (Operation Specimen) Showing Massive Secondary Deposits of Carcinoma Four Years After Excision of the Rectum.—Mr. W. B. Gabriel.

(1) Pseudo-membranous Colitis Due to Carcinomatous Obstruction of the Colon. (2) Unusual Bezoar of Terminal Ileum.—Mr. A. Dickson Wright.

Scald Stricture of the Sigmoid Colon.—Mr. Henry Thompson.

Four Pathological Problems.—Dr. Cuthbert Dukes.

Crohn's Disease with Ileo-rectal Fistula.—Mr. E. G. Muir.

Atypical Ulcerative Colitis in an Infant Requiring Colectomy.—Mr. F. R. Sutton (for Mr. E. G. Muir).

Colon of a Female Aged 26 Showing Familial Polyposis and a Carcinoma with Metastases in the Regional Lymph Nodes. Total Colectomy: Fulguration of Rectal Polyps: Ileo-rectal Anastomosis.—Mr. Ronald W. Raven.

Tuberculosis of Anus Cured by Local Streptomycin.—Mr. J-J. Brossy (for Mr. M. R. Ewing).

[*February* 10, 1954]

Simplified Treatment of Fistulæ in Proctology

By G. B. Ettore Simonetti, M.D.(Milan)

Anatomical Data

Santorini (1739) was the first to make a detailed description of the anal musculature, and of the pelvic diaphragm, splendidly illustrated by V. d. Meer. His studies were continued by Girardi (1775), Kohlrauch (1854), Béraud (1857), Holl (1897) and more recently by Lloyd-Davies (1936), and Levy (1936). The complications involved in operating on the ano-rectal areas have led many authors carefully to extend their anatomical studies in relation to surgical techniques.

I mention only briefly the important workers of St. Mark's Hospital School, among whom are Milligan and Morgan (1934, 1935), Milligan (1943), Dukes (1944), Gabriel (1921, 1948), and Morgan (1949).

With regard to the simplified technique which I use, only one point seems to me to be worth mentioning, namely, the arrangement of the fibres having a longitudinal and circular direction in the sphincter musculature, forming a "web-like arrangement" from which a functional complex derives, so that the linear longitudinal resection does not imply the spontaneous retraction of the resected surfaces, as in the case of muscles with fibres having a homogeneous direction, and they practically remain in close contact.

Classification

In order to describe all kinds of fistulæ met with in proctology by simplified terms, I use a classification which divides the fistulæ into two groups:

(*a*) *Subcutaneous* and *submucous* fistulæ which do not involve the sphincter ani.

(*b*) *Sphincteric* fistulæ involving the sphincteric musculature. These, in turn, can be divided into:

(1) *First degree fistulæ* (low-level anal fistulæ according to Milligan and Morgan, 1935) entering the anal canal at the septum intermusculare, and between the sphincter ani externus subcutaneus, and the lower border of the sphincter ani externus superficialis.

(2) *Second degree fistulæ* (high-level anal fistulæ according to Milligan and Morgan) involving the sphincter ani externus profundus, and situated below the levator ani.

(3) *Third degree fistulæ* extending beyond the levator ani (ano-rectal fistulæ, according to Milligan and Morgan).

The diagram shown in Fig. 1, which I use in my case sheets to record results of the rectal examinations and proctological treatments, makes it easy to mark the course of the various tracts.

Personal Technique

In the mechanism of fistula formation and their persistence or complications, physical laws prevail; first of all that of gravity which in the case of hindered drainage will cause the formation of a septic fistulous sinus, in which the purulent fluids stagnate. These are subject to Pascal's principle, just as any fluid, particularly when, as is often the case, a temporary obliteration of the drainage openings occurs.

An experience of over twenty years in proctology, as well as the observation of a great number of cases treated in different ways, has led me firmly to believe that the one condition essential to the cure of fistulæ, and which should be regarded as a law for the successful proceeding of the treatment, whatever the method used, is that *the circularity of the tract should be discontinued on the whole of its length, and that a new level of free drainage should be brought about, "towards the intestinal lumen, and outwardly", both longitudinally and perpendicularly to the axis of direction of the tract, with an outward and downward declivity* (Fig. 2).

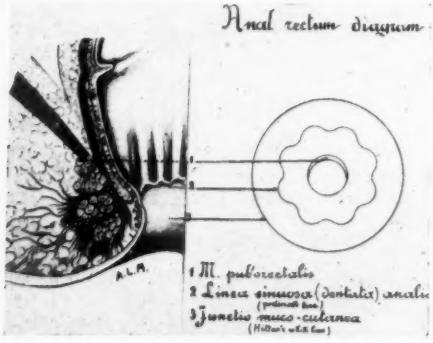

FIG. 1.

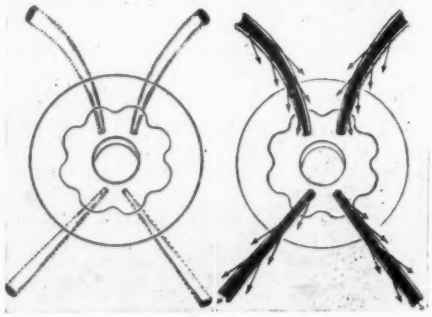

FIG. 2.

In practice some concessions must be made in the geometrical strictness of that law; however, fundamental disregard of it will lead to therapeutic failure. In order to eliminate the fistulous tract, it is essential to remove any hindrances to the free drainage to the exterior, and then to resect all the tissues lying between the bottom of the canal and the exterior in the case of subcutaneous fistulæ, and between the bottom of the canal and the intestinal lumen, including the sphincter as well, in the case of sphincteric fistulæ.

For that purpose I use diathermic resection by means of a steel wire used as an active electrode with bi-polar current and indifferent dorsal electrode. Thus I am able to get a straight cut from within out at a right angle, on the sphincteric fibres, and with no loss of blood, and with free drainage. When the surgeon is sufficiently experienced the intervention can be performed in one stage, with the patient in a knee-elbow position, after local anæsthesia. The application of the steel wire electrode like a snare makes it possible to obtain a clamp action, with successive approach and mechanical ischæmia of the tissues to be cut, and then an increase of the hæmostatic power of the diathermic incision.

Any secondary sinuses can be removed by repeating the operation, as well as any protuberance fairly frequent in the superficial planes since these might give rise to healing bridges. The wound will heal spontaneously *per secundam*, with a soft, elastic even scar. *No packing* should be inserted into the wound as this might cause the falling apart of the resected surfaces, with the danger of loss or impairment of control. It is imperative that the cut should be thoroughly hæmostatic, yet not too coagulating lest hæmatomas or too widespread eschar formations should occur favouring the falling apart of the resected surfaces, and, therefore, defective cicatrization, harmful to the control. Healing takes from 20 to 40 days during which any untreated offshoots which might interfere with healing can be removed. These may be identified through small hæmorrhages with local, purulent secretions in some parts of the main wound. It will then be possible to proceed to their resection by means of a second intervention much easier than the first, so that complete healing is delayed only a few days.

The method is highly successful as the careful clearance ensures healing for every case of fistula. Its realization, however, is most exacting, as it requires a perfect mastery of the electric bistoury.

The Sphincteric Fistulæ from a Functional Aspect

First degree fistulæ are most frequently encountered; however, second and third degree fistulæ are also possible. In any case it is interesting, particularly in the case of complicated tracts, to have

most simple clinical data to differentiate between the functional area belonging to the voluntary musculature involved by the tract and, therefore, to be resected for therapeutic purposes, and the area belonging to the residual voluntary musculature. It is sufficient to introduce a probe into the tract, and a finger into the sphincter at the same time, asking the patient to thrust as he would do in the act of defæcation. In that way the contraction of the muscles medially situated to the tract can be easily felt, as well as a thrust of the probe on the finger which is all the stronger as the functional area of the voluntary musculature situated beyond the fistula is wider, and, therefore, not likely to be involved.

This procedure which I use in the systematic examination of sphincteric fistulæ gives a fairly correct idea of the voluntary function of the unaffected area.

Statistical Data

In 1,021 cases of fistulæ treated by me over a period of twenty years, I found 139 cases (13·61%) of subcutaneous and submucous fistulæ (89 pilonidal sinus, 41 subcutaneous, from infections of the external pole of the fissures of the posterior or anterior commissures, ano-vaginal, ano-perineal, and ano-scrotal, and 9 submucous from cryptitis), 665 cases (65·13%) of first degree sphincteric fistulæ, generally from infections of the inner pole of fissures or superficial abscesses, 141 second degree sphincteric fistulæ (13·81%) from infections of the ischiorectal fossa, 76 third degree sphincteric fistulæ (7·44%), generally from pelvi-rectal infections, or as a recurrence following the usual interventions. Of the cases treated 55% were male and 45% female.

In every case it was possible entirely to remove the fistulous tracts, generally at one operation, and in 10% of cases with two or more interventions. Loss, or appreciable impairment of the control, was never observed.

Figs. 3–6 are descriptions of some of the more interesting cases treated by me recently.

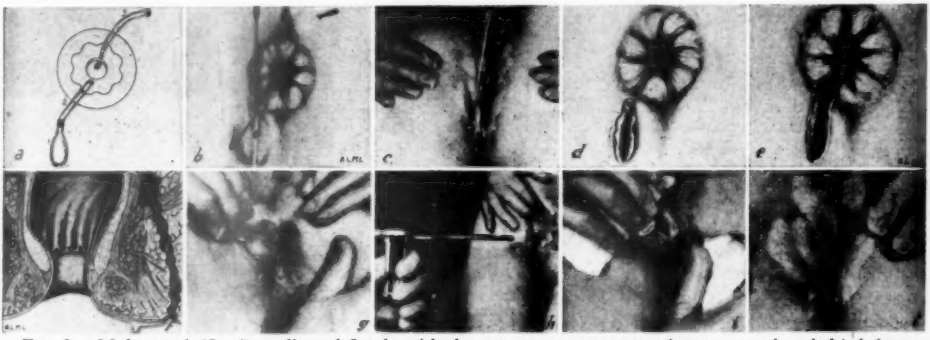

Fig. 3.—Male, aged 65. Complicated fistula with three tracts respectively of first, second and third degree (a, b, c). Treated on 15.12.53 (d, e, f, g). Dressing of the wound with no packing (h). Immediate, perfect control. Falling off of the eschar one week later (i). Healing completed in 30 days (l).

Fig. 4.—Male, aged 45. Horse-shoe fistula (a, b). Previously operated upon elsewhere four times by the usual methods with no success. Treated by means of two distinct diathermic cuts on 7.1.54 (c, d). Immediate, perfect control. Healing completed in 27 days (e).

Conclusions

The method of diathermic fistulotomy which I have just described affords the conditions which are essential to the cure of fistulæ (opening of the tract with adequate external drainage), while adhering to the recognized measures for the maintenance of control (incision of the sphincteric fibres at a right angle, with no packing whatever so that the margins of the wound can heal without loss of continuity).

Diathermic fistulotomy:

(*a*) Makes it possible to take advantage of slow resection by the old ligation method which was recently resumed and widely used, particularly in France, with promising results (Kaufmann, 1936; Arnous, 1948; Henry, 1952); but without its limitations (prolonged treatment, pains, &c.). It also makes possible the advantages of the common fistulotomy and fistulectomy, and combined fistulotomy and fistulectomy (Aronsson, 1948) but without loss of control.

(*b*) It sterilizes the tissues and is followed by lymphorrhoea, a natural drainage and a protective barrier against infection.

(*c*) It can be done with a local anæsthetic. There is no blood loss; dilatation is not required.

FIG. 5.—Female, aged 22. Pilonidal sinus (a, b). Previously operated upon elsewhere four times with no success. Treated on 15.1.54. The case reveals multiple tracts extending beyond the area normally excised by elliptical excision (c, d). Healing completed in 40 days (e).

FIG. 6.—Male, aged 52. Operated upon once elsewhere with no success. Normal examinations reveal only one external subcutaneous tract (a). In the course of treatment 4 more tracts were revealed (b, c, d) one of which is a sphincteric tract. Treated on 3.2.54. Healing completed in 30 days (e).

(*d*) It is precise since it renders possible the investigation and detection of any offshoots from the main tract which may have escaped previous examination, even radiological. This examination is made in a blood-free field, uncluttered with instruments.

(*e*) It is possible to repair in a second or successive stage the not too frequent cases of complicated fistulæ in which some secondary tracts might have escaped the primary treatment.

(*f*) Usually the patient is ambulatory and need not be hospitalized. The post-operative course is usually uneventful.

However, I cannot close this brief report without stressing once more that while the method is highly satisfactory the operation requires considerable skill and experience in the performance of diathermic resections.

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Complete Rectal Prolapse Following Removal of Tumours of the Cauda Equina. Two Cases

By E. C. B. Butler, F.R.C.S.

Both these patients were young women and their case histories present certain features which may have a bearing on the ætiology, treatment and post-operative complications of this distressing condition.

Case I.—Girl aged 18, admitted to the London Hospital in January 1948.

In 1942 Mr. D. W. C. Northfield had removed an ependymoma from the cauda equina. There had been no local recurrence. Following the operation the girl showed signs of a partial lesion of sacral nerves 1–4. There was a saddle area of anæsthesia over the buttocks and in 1948 one of her ankles had been arthrodesed because of a flail joint. Since her operation she had suffered from urinary incontinence and for the past five months her rectum had prolapsed.

On admission her general condition was good but she was in considerable distress from double incontinence. Despite her large prolapse voluntary contraction of the anal sphincters was quite good.

18.2.48: Rectosigmoidectomy was carried out from below and about 6 in. of bowel was removed.

26.2.48: Severe secondary hæmorrhage occurred from the suture line; this was treated by irrigation of the rectum, packing the bowel and an emergency left inguinal colostomy.

7.4.48: The colostomy was closed.

5.3.50: Admitted with a large recurrent prolapse; in other respects her condition was unchanged.

7.3.50: Lithotomy Trendelenburg position. Laparotomy showed the pelvic colon running down to the distal rectum surrounded by a deep pouch of peritoneum which extended between the colon and vagina and also part way behind the colon on the right side.

The whole of the hernial sac was removed from above; the colon was drawn up and anchored to the side wall of the pelvis by interrupted silk sutures; a raw surface was thus left between the colon and vagina anteriorly and the sacrum posteriorly. The peritoneal floor was reformed at a high level and ventro-fixation of the uterus carried out. Since that date she has had no more rectal prolapse and her control is good. Her urinary incontinence improved a great deal after her second operation and she now only gets trouble if she coughs or sneezes.

Case II.—Woman aged 29, admitted in 1953.

In 1947 a very large ependymoma of the cauda equina had been excised by Mr. D. W. C. Northfield; it weighed 45 grammes. Since that date she had suffered from continuous urinary incontinence but this had been greatly relieved by repeated endoscopic resections of the bladder neck carried out by Professor V. W. Dix.

For the past year she had complained of increasing rectal prolapse.

On examination she was in a miserable state; mentally very depressed by her long illness and by the presence of a huge prolapse.

There were signs of gross lesions to her sacral nerves with absent ankle-jerks; saddle anæsthesia and very poor sphincteric tone.

Over the sacrum was a scar representing the site of a large trophic ulcer which had been excised.

For the past few months she had not gone out because of her prolapse and she was in every way a more serious problem than the first girl.

11.2.53: Lithotomy Trendelenburg position. Laparotomy showed a deep hernial sac between the rectum and vagina. The rectum and pelvic colon were completely mobilized and the hernial sac was excised. The mobilized bowel was then drawn out through the anus and a rectosigmoidectomy performed by my senior registrar Mr. D. V. Evans. About 2 ft. of bowel was removed. The peritoneal floor was repaired from above and a ventro-fixation carried out.

19.2.53: There was a sudden severe secondary hæmorrhage from the suture line; this was treated by irrigation and a temporary transverse colostomy.

25.3.53: The colostomy was closed.

Since that date she has remained well; her mental state has returned to normal and she is able to lead an ordinary life. It is, of course, too soon to be able to claim any permanent success from this operation but the immediate results are gratifying.

Comment.—In both these cases it seems probable that the prolapse was due to a partial paresis of the levator ani muscle following an operation on the cauda equina which produced, in both women, a partial lesion of the sacral plexus.

Similar prolapses are occasionally met with in young women with spina bifida occulta, when concurrent uterine prolapse may also be present.

It was difficult at operation to define the middle portion of the levator ani but one could establish that the condition was a large sliding hernia between the rectum and vagina. The findings at the second operation on Case I show clearly the reason for the high recurrence rate following perineal rectosigmoidectomy; only the lower part of the large hernial sac is usually removed so that further prolapse is often inevitable despite strengthening of the levator muscles from below.

A combined abdominoperineal rectosigmoidectomy, although a formidable procedure, seems to be well tolerated even by elderly patients and this operation appears, at present, to offer the best chance of a cure, as in addition to the removal of all the excess bowel, the hernial sac is excised and the pelvic floor strengthened.

In these young women no attempt was made to reinforce the levator muscles from below although this is usually done in older patients.

Thorough removal of the sac and complete mobilization of the bowel leaves raw areas in front and behind the colon and the resulting fibrous adhesions at these sites are probably important factors in the prevention of a recurrence.

Unabsorbable sutures between the colon and the side wall of the pelvis are valuable for a time as they help to anchor the bowel until adhesions have formed; it is doubtful if they in themselves are of any permanent value.

Ventrofixation of the uterus is useful since it helps to hold up the posterior vaginal wall and through it the anterior part of the adherent rectum.

Both the cases quoted above had severe secondary hæmorrhage from their suture lines which in Case I might well have proved fatal. It is curious that this complication should have arisen since I have not met with it in anterior resections for carcinoma.

Table I gives the results to date of 10 cases of complete prolapse treated by surgery; 5 have had combined rectosigmoidectomy, the longest follow-up is two years and there have been no recurrences. 7 had intra-abdominal repair only, without excision of the redundant bowel; the longest follow-up is five years, 2 have recurred and required further operation. Both perineal operations relapsed.

TABLE I.—COMPLETE PROLAPSE OF THE RECTUM

Age	Cause	Operations	Date	1954
20	Cauda equina tumour	R.S.	1948	
		A.R.-V.-P.	1950	Well
30	S. bifida occulta	A.R.-V.-P.	1947	Well
67	—	R.S.	1948	
		A.R.-V.-P.	1950	Well
49	—	A.R.-V.	1949	Well
49	—	R.S.	1949	
		A.R.-V.	1951	
		C.R.S.	1953	Well
65	—	A.R.-V.	1949	
		P.	1951	
		C.R.S.	1953	Well
62	—	A.R.-V.	1952	Well
64	—	C.R.S.	1952	Well
29	C. equina tum.	C.R.S.	1953	Well
53	—	C.R.S.	1953	Well

R.S., Rectosigmoidectomy. V, Ventro-fixation.
P, Repair of the perineum.
A.R., Intra-abdominal repair without excision of gut.
C.R.S., Combined rectosigmoidectomy plus abdominal repair.

These results make me believe that combined rectosigmoidectomy with repair of the perineum from below and excision of the hernial sac from above offers the best chance of a cure in severe cases. In elderly women who are unfit for major surgery pararectal injection of about 200 c.c. of sterile castor oil combined with a Thiersch's operation often gives great relief and is well worth a trial.

Some Technical Aspects of Anterior Abdominal Resection for Carcinoma of the Rectum and Other Lesions of the Rectum and Rectosigmoid

By C. PATRICK SAMES, M.S., F.R.C.S.

OPINION regarding the wisdom of sphincter-saving operations for carcinoma of the rectum and rectosigmoid is still crystallizing, and, although receiving increasing pathological support, may be judged to have had a severe set-back by the high incidence reported of local implant at or about the suture line. However, the recent resurgence of interest both in this country and the U.S.A. in such operations for carcinoma has led to wider familiarity and to increasing application for non-malignant conditions, especially diverticulitis of the lower sigmoid. Especially is this the case where complications have occurred such as pericolitis and cicatricial narrowing, the so-called Graser's tumour. Other non-specific granulomata and endometriosis are likewise amenable to such treatment.

I believe that anterior abdominal resection is the operation of choice where conservation of the sphincter is aimed at, and is preferable to "pull-through" operations or abdomino-sacral resection, the chief objection to these methods being the high incidence of lowered rectal sensibility which follows them.

In addition to the fear of local implant in cases of anterior resection for malignant disease, there is the added risk of stricture and fistula formation irrespective of the pathology for which the operation is undertaken, together with the possibility of local or general peritonitis and local pelvic cellulitis. I maintain that careful attention to technical detail can minimise the complications, the aim being to procure healing at the suture line by what is virtually first intention.

Colostomy or No?

I prefer, wherever possible, to avoid colostomy except in cases of severe obstruction or perforation, and do not even make use of cæcostomy as a safety-valve at the time of operation. The passage of solid fæces through the suture line is the best dilator and physiological preventative against stricture formation. That this was practised before the days of local and systemic chemotherapy by such surgeons as Pannett speaks highly for their surgical craftsmanship. When colostomy is imperative it should be in the transverse colon, through a small right transverse rectus incision, taking care to pull the colon as far to the left as possible, and thus making the vent well to the patient's right. Ample colon is thus provided should the second operation entail mobilization of the splenic flexure for more extensive diverticulitis or a flush tie of the inferior mesenteric in carcinoma. No "gun barrelling" of the colon is indicated, and the transverse incision permits of easy fashioning of a skin bridge between the divided ends of the colon. Closure of the colostomy should be as soon as possible, preferably within two or three weeks. My experience of extraperitoneal closure has been so disappointing that I have abandoned it and make a formal intraperitoneal closure now in all cases and, as yet, have had no cause for regret.

Pre-operative Preparation

I rely on phthalylsulphathiazole cream for intestinal antisepsis given usually by mouth, but also into the distal loop when a colostomy has been previously performed. I prefer to keep streptomycin for the day of operation, and the subsequent three to four days, rather than run the risk of establishing resistant organisms before operation. However, I have used the broad spectrum antibiotics pre-operatively when local inflammatory masses have been present in the left iliac fossa in order to hasten resolution.

I prefer to dispense with pre-operative washouts, except in cases with a colostomy, or where subacute obstruction is judged to be present. It is far easier to deal with formed fæcal masses at operation than with the fluid results of an unsatisfactory washout.

Preliminary packing.—I prefer always to suture the bowel around a soft rubber tube of adequate lumen (1·5 cm. diameter) yet prefer to have completed the back row of sutures before pulling the tube through. Therefore, before commencing the operation the rectum is packed with a two-foot length of 6 in. folded gauze, to the end of which is firmly secured a length of the selected rubber tubing (Fig. 1). The patient is placed in the "jack-knife" position, and, after forceful stretching of the sphincters, the gauze, soaked in 1 in 1,000 silver nitrate solution, is closely packed well up to the suspected lesion, and the first part of the rectal tube just engaged within the anus. When the time comes, traction on the gauze is quickly followed by the appearance of the tube. Fergusson's speculum and vaginal packing forceps are ideal for this procedure.

Abdominal incision.—This should be a left paramedian and adequate. However, excessive length only makes small bowel control more difficult. Extension upwards may be required if mobilization of splenic flexure is resorted to. A lateral extension with division of the anterior and posterior rectus sheaths, but leaving the muscle intact, is of great help when difficulty is experienced in reaching the splenic flexure.

Site of proximal section.—The pelvic colon and upper rectum are mobilized in the usual way as for routine abdomino-perineal excision. The anatomy of the vascular pattern in the mesentery of the sigmoid and rectosigmoid has been adequately dealt with by Goligher (1949), but it must be frankly admitted that even with the aid of transillumination the mesentery in cases of diverticulitis may be so fat-laden as to preclude accurate assessment and the point of proximal section must be found by trial and error. It is, therefore, wise to tie off the inferior mesenteric, and make the bowel section early in the proceedings, and, after spending time mobilizing rectum and pelvic peritoneum, to return to it to check on its viability. The bowel, of course, should be sectioned obliquely away from the mesenteric border, so as to give adequate lumen, and ensure good blood supply to the anti-mesenteric border. It need hardly be emphasized that in cases of carcinoma the surgeon must not be influenced in any way by the length or mobility or otherwise of the pelvic loop, and must be prepared to mobilize the splenic flexure or perform a flush tie, and even to abandon hope of restorative resection. It cannot be over-emphasized that sphincter conservation is *not* synonymous with conservative resection except in cases judged to be frankly palliative.

Site of distal section.—The rectum must be adequately mobilized before suturing is attempted; posteriorly to the fibrous septum extending forward from the sacro-coccygeal joint, laterally after the division of the lateral rectal folds and anteriorly to below the post-vaginal vault or the seminal vesicles. In cases of carcinoma, as soon as the lesion has been adequately mobilized it is best to apply a soft clamp below in the hope of preventing any intraluminal spill. Adequate mobilization results in a surprising length of rectum by undoing the anteroposterior curve in the sacral hollow, and, to a lesser degree, the sigmoid curve. Gentle traction no doubt results in tenting upwards of the perineum and anal sphincters.

On the posterior and lateral walls of the middle part of the rectum will be found the terminal divisions of the inferior mesenteric vessels (now the superior hæmorrhoidal). These vessels numbering

2, 3 or 4 are underrun with an aneurysm needle or Cleveland ligature carrier and divided (Fig. 2) just below the selected line of suture. This "double ligation" of the vascular pedicle prevents troublesome reflux bleeding—especially of veins, and facilitates the suturing. The vessels are slightly shorter than the rectum they supply, and their division, together with the surrounding fibro-fatty mesentery, results in further unfolding of the sigmoid curve and consequent lengthening. Every millimetre gained is of assistance, especially in the small narrow male pelvis.

In lesions above the peritoneal shelf it is tempting but, I believe, unwise to perform a purely intraperitoneal suture without rectal mobilization. This would only be safe provided one is sure of the innocent nature of the pathology, and can resect through the vascular arcades leaving the inferior mesenteric artery intact. However, the inflammatory masses associated with diverticulitis seldom permit this and there are always the unsuspected cases of dual pathology—co-existent carcinoma and diverticulitis, to be considered.

FIG. 1.—Gauze packing with attached rectal tube and Fergusson's forceps for preliminary packing.

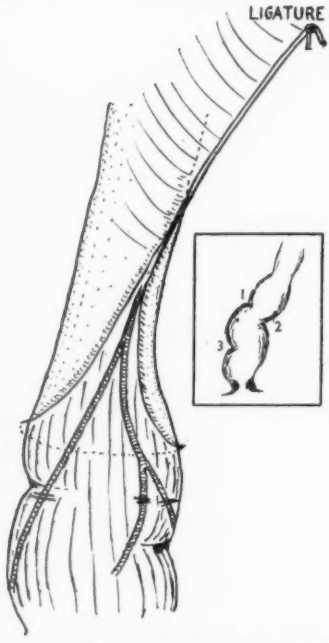

FIG. 2.—Mobilized rectum showing site of secondary ligation of superior hæmorrhoidal vessels (marked =) to prevent reflux and help unfolding of sigmoid curve of rectum. Inset shows sigmoid curve of rectum with corresponding valves of Houston 1, 2 and 3.

The anastomosis.—This is undertaken with interrupted No. 40 cotton sutures throughout. After careful packing of the pelvis the proximal colon (now checked for its viability) is gently approximated by three posterior sutures. These are applied before final section of the rectum, as in total gastrectomy. The rectal wall with its friable outer longitudinal muscle coat and no serosa closely resembles the œsophagus for suturing. These three sutures are therefore placed horizontally to prevent cutting out, and no reliance is placed upon them other than for approximation. The rectum is then cut across with scissors, and some of the contained gauze packing cut off and discarded. The posterior layers are then carefully and accurately sutured with interrupted sutures—about one suture to every 2 to 3 mm., taking meticulous care to pick up all coats, and get accurate inversion of the mucosa. After completion of this layer, the gauze is then pulled on and the tube, previously placed in the rectum, comes readily to hand. The gauze is severed from its connexion with the tube, and the latter gently eased into the colon for a matter of 7 to 10 cm. Much fumbling by the assistant beneath the drapes is thus avoided. Although the colon has been cut obliquely, to ensure an adequate lumen and prevent stricture, I make a routine "T" extension on the anti-mesenteric border. The anterior layer is completed by a series of sutures, looped on the mucosa, to ensure accurate inversion, with a few further approximating mattress sutures disposed transversely. The peritoneal floor is reconstructed around the colon leaving the anastomosis well below in the extraperitoneal plane—a compensation for

having a suture line one side only of which is lined with serosa. Drainage of the pelvis is afforded by a rubber tube disposed retroperitoneally, and coming out through a stab wound rather far out in the left iliac fossa, thus minimizing the risk of small bowel obstruction.

I have never thought it necessary to rotate the proximal colon before suturing, as is recommended by some authorities.

The suture line can usually be felt at about 6 to 7 cm. (sometimes 5 cm.) from the anal verge. It is usually disposed somewhat to the left, and, although palpable, cannot, as a rule, be adequately palpated by the finger. The post-operative recovery is remarkably smooth, there being in my experience far less shock and post-operative morbidity than in abdomino-perineal resections. There seem to be no post-operative bladder troubles, due, perhaps, to the fact that there is a new "rectum", which prevents backward dislocation of the bladder in the hollow of the sacrum. Discharge from hospital on the twelfth to fourteenth day is the rule rather than the exception.

When I first commenced to practise this operation, I taught that every case developed a fæcal fistula for a variable time, but with practice I find that such a complication is an exception. I believe that attention to detail, especially as to suturing, results in healing by first intention. Experimental work by Vink (1954) in Amsterdam tends to show that intra-luminal implant of cancer cells in the bowels is more probable in bowels rendered "antiseptic" by chemotherapy, than in colons with normal flora. There may come a time when we have to return to colon surgery without these ancillary aids, and so it behoves us to pay attention to these other details.

The avoidance of morbidity in anterior resection of the rectum can thus be summarized under the following headings:

(1) Meticulous, accurate and painstaking suturing.
(2) Avoidance of over-suturing.
(3) Avoidance of tension by proper mobilization of the parts.
(4) Avoidance of temptation to anastomose above the peritoneal shelf.
(5) Avoidance of clamps or diathermy to distal end.
(6) Get formed fæces passing through suture line as soon as possible.

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The White Line of Hilton

By M. R. EWING, F.R.C.S.

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JOHN HILTON was appointed assistant surgeon to Guy's Hospital in 1844 (Plarr, 1930). This must have been regarded as a somewhat unusual appointment, for he had served no apprenticeship in surgery, either to the hospital, or to any member of its staff. He had, in fact, passed the greater part of the sixteen years since graduation in the dissecting rooms of the medical school, a fact which had earned for him the nickname "Anatomical John".

In 1860, '61 and '62 he gave in the College a course of close on 40 lectures, which appeared separately in the *Lancet* soon after their delivery. They must have been warmly received, for in 1863 Hilton was induced by the "urgent solicitations of my professional brethren" to collect them together and to publish them as a single volume entitled "On the Influence of Mechanical and Physiological Rest in the Treatment of Accidents and Surgical Diseases and the Diagnostic Value of Pain". This clumsy title was shortened in the second edition in 1877 to "On Rest and Pain" and it is by this name that it has become widely known and accepted as one of the classics of surgical writing.

In the 12th of these lectures (Hilton, 1877), he discussed the comparative insensibility of the upper portion of the mucous membrane of the rectum, and the contrasting "great sensibility, difficult dilatation, and enduring power of contraction, the characteristic of the lower portion of rectum". He then went on to consider some points in the anatomy of the lower rectum and anal canal and drew attention to what he believed to be a very important point in surgical anatomy, viz. a white line in the anal canal which can serve as a valuable guide to the surgeon when he aims to cut the sphincter muscle, as, for example, in the relief of the pain of an anal fissure.

Of this line he writes "... a white line, which in the living subject any surgeon can recognize, shows the junction of the skin and the mucous membrane. That white line corresponds exactly with the lineal interval between the external and internal sphincter muscle. It is an important landmark, exact and truthful so that it can be relied upon". Later in his lecture he goes on to say that the special object of the engravings which illustrate his text is "... to show the precise line of junction or demarcation of the internal and external sphincter of the anus, and that line exactly corresponds (I have tried it several times) with the white line of junction between the skin and the internal mucous membrane".

As far as I am aware, he made no other communication on the anatomy of the rectum and anus, but on the strength of what is really a very short and almost casual reference introduced only to sustain his argument, his name has since become perpetuated in this connexion in authoritative anatomical and surgical teaching, as "Hilton's line" or perhaps more often as "Hilton's white line".

I must confess, however, that I have never been quite clear what is meant by Hilton's line and although his original description seems definite enough, I have never found it to be the precise anatomical feature which his writings suggest. Looking round for help, I first turned to the standard anatomical textbooks. In Cunningham (Cunningham, 1951) the lining of the anal canal is described as consisting of 3 areas. First, a cutaneous zone round the margin of the anus, possessing hair follicles and glands. This merges very gradually above into a smoother, hairless area still lined with squamous epithelium. This, in turn, gives way at an abrupt sinuous line at the level of the anal valves to the mucous membrane covering the lowest part of the anal columns. This abrupt line is here called the "white line of Hilton", although it is better known as the dentate or pectinate line. I was surprised to learn that Hilton's line was thought to be at this level. The valves are much more obvious in the cadaver than in the living subject, and Hilton must surely have demonstrated them many times, in the course of his long experience as a teacher of practical anatomy. I find it hard to believe that had he meant the line of the valves he would not have said something of their very distinctive, sinuous and dentate disposition.

In the 30th edition of Gray's Anatomy (Gray, 1949) we read that the lower half of the anal canal is lined with skin and that the junction of skin and mucous membrane is indicated by a white line. There is no mention of Hilton. Knowing that "Gray" has for the past twenty years been edited from Guy's, I was inclined to believe that this failure to commemorate the name of Hilton, who was after all one of their earliest anatomical and surgical teachers, was the result of the not unnatural fear of the ambiguity which use of his name might occasion: but I am afraid there must be other reasons, for not even the subsartorial canal is allowed a parenthetical "Hunter".

Next I turned to the 8th edition of Buchanan's Anatomy (1949). The pectinate line is here taken to mark the junction of mucous membrane and skin. At a distance varying from 3 mm. to 9 mm. below is Hilton's white line. This latter is not defined further, save for a remark about its colour. The portion of the anal canal between these two lines (the pectinate above and Hilton's below) is lined with squamous epithelium transitional between the glandular epithelium of the upper part of the canal and the true skin at the anal orifice below. This zone is called the pecten.

This description follows, in the main, that usually given in the standard American textbooks, with the further addition that the term pecten is there almost always coupled with that of Stroud. He was a zoologist at Cornell who in 1896 (Stroud, 1896) published an interesting paper dealing primarily with the comparative anatomy of the anal papillæ. In the course of this contribution he used the term pecten—and, as far as I can ascertain, he was the first to use it—to describe the area which he found in all mammals and which marks the transition between the mucous membrane and the skin. He drew attention to the following features of this zone of skin (1) it is smooth and shining, (2) its colour is intermediate between that of the mucous membrane and the skin, (3) it has no sweat glands, (4) it is covered by modified stratified epithelium, (5) it is bounded above by the dentate line and below by the junction of the external and the internal sphincters and after this, in parenthesis, he put "Hilton's white line."

Here again is mention of Hilton's line but with no attempt to define it other than to place it 3–9 mm. below the dentate line. It seems perfectly clear that there was no doubt in Stroud's mind that the dentate line is quite distinct from Hilton's line which he took to indicate the line of junction of the two sphincters.

In the earlier editions of Cunningham, in which Hilton's line and the dentate line are made to coincide, I had found the alternative name of the ano-cutaneous line of Herrmann. He was a French anatomist who in 1880 published a most detailed account of the histology of the lining of the anal canal. The chief interest of this contribution for me was in bringing to my notice a paper by two of Herrmann's teachers, Robin and Cadiat, which had appeared in the same journal six years earlier (Robin and Cadiat, 1874). I found here a description of the minute anatomy of this region which I believe for clarity and accuracy puts to shame most of what one finds on the subject in the standard textbooks of to-day.

However, before I confuse the issue any further, I think that I should go back to restate the essential features of Hilton's line, as laid down in his original description.

Here he made the following points about his surgical land-mark: (1) it ". . . corresponds exactly with the lineal interval between the external and internal sphincter muscle". (2) it marks the junction between skin and mucous membrane, (3) it is ". . . exact and truthful, so that it can be relied upon", and ". . . in the living subject any surgeon can recognize (it)", (4) it is a line, (5) it is white.

My argument is that these points can properly be amended. The first I will allow; the others should be read in the negative.

Fig. 1 is a reproduction of the beautiful lithograph from the paper by Robin and Cadiat which shows the minute structure of the wall of the anal canal when it has been split longitudinally. One can readily recognize the junction between the internal sphincter and that part of the external sphincter which lies in the same plane and which we now call "subcutaneous". The lower margin of the internal sphincter is smooth, rounded and clearly defined—there is no mistaking it. One can in any longitudinal section of the anal canal readily, confidently and precisely indicate this level. I have satisfied myself on this score many times (Fig. 2).

And not only can one be sure of it in stained sections: it is a level which can be detected just as confidently during life, especially when the sphincters are in spasm, as, for example, in the presence of a painful fissure. Milligan and Morgan (1934) in their classical surgical-anatomical study of twenty years ago pointed out that this interval is marked clinically by a slight depression 3–6 mm. ($\frac{1}{8}$ in.–$\frac{1}{4}$ in.) in breadth felt immediately under the skin just inside the anal verge.

But it is a level whose easy detection (without dissection) is dependent on a functioning sphincter, whether this be normal tonic or a pathological spasmodic contraction. I am satisfied that this was the level that Hilton meant. He says so pointedly, and I believe so significantly ". . . a white line which *in the living subject* (the italics are mine) any surgeon can recognize." Here is a level which he felt

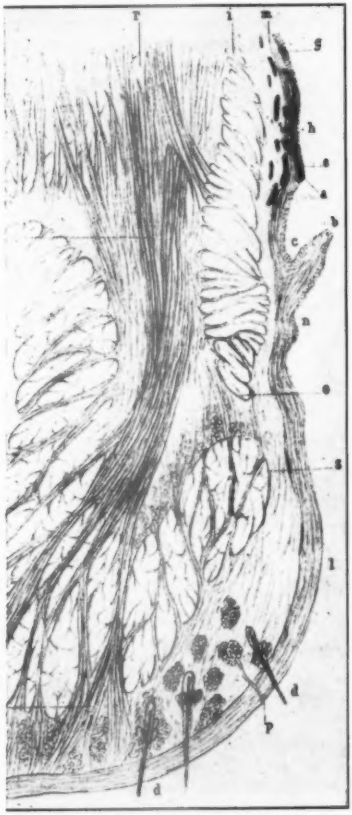

FIG. 1.—Longitudinal section of anal canal. The only relevant letterings are: a = junction of squamous and columnar epithelium. b = anal valve. o = lower margin of internal sphincter. (Modified from a lithograph in Robin and Cadiat (1874) *J. Anat., Paris*, **10**, 589.)

FIG. 2.—Longitudinal section of anal canal taken at post-mortem. H.E. (× 4). The position of the anal valves and the depression which marks the interval between the adjacent margins of the internal and external sphincters are clearly seen.

he could indicate with confidence, and the accuracy of its placing he could confirm, as readily by operation on the living subject, as by dissection in the dead one ". . . exact and truthful and can be relied upon."

If we accept, then, that the intersphincteric level, or the anal intermuscular septum (Milligan and Morgan, 1934) as it has now become widely known in this country, indicates the position of Hilton's line, does it also satisfy the second requirement of marking the junction between mucous membrane and skin?

Using special stains for mucin I have established in many sections taken at post-mortem that the line of junction of squamous and of columnar epithelium is usually about 1 cm. above the lower

margin of the internal sphincter (Figs. 3 and 4) and may even be above the line of the valves (Fig. 5). The lower margin of the internal sphincter descends almost always to a level varying from 6–12 mm. below the dentate line and embraces all of that zone lined with modified skin, which is variously called, by Stroud the pecten, by Cunningham the intermediate zone and by Robin and Cadiat and by Herrmann as the "zone cutanée anale lisse". I must admit, however, that there is so much interpenetration of the epithelia (Harris, 1929) that it is often exceedingly difficult in a histological section to mark precisely the line of junction between the two. Bearing this in mind, it should surely occasion no surprise to learn that when we guess this level clinically, as, no doubt, did Hilton, our estimate might be wrong by as much as 1 cm. or more.

Assuming then that we are right in believing (1) that Hilton's line is the interval between the internal and the external sphincters, and (2) that he was wrong in believing that the change-over from mucous membrane to skin occurs at this level, is the intersphincteric interval marked by a *white* line? For, after all, that is what Hilton called it and what posterity has continued to call it. In his engravings it is marked clearly as a precise white line measuring only a few mm. across. I have examined this area closely both in the living subject and in the cadaver and I cannot claim that I have ever seen such a definite line as features in his illustrations (Fig. 6).

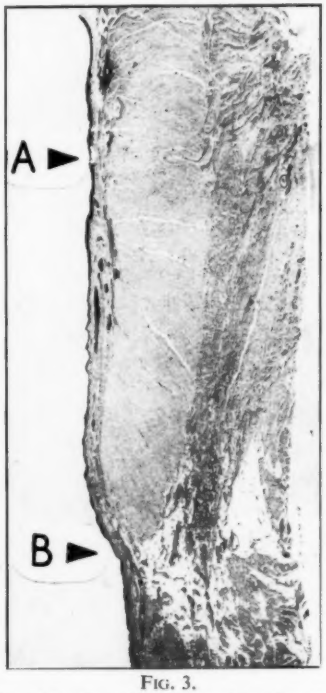

FIG. 3.

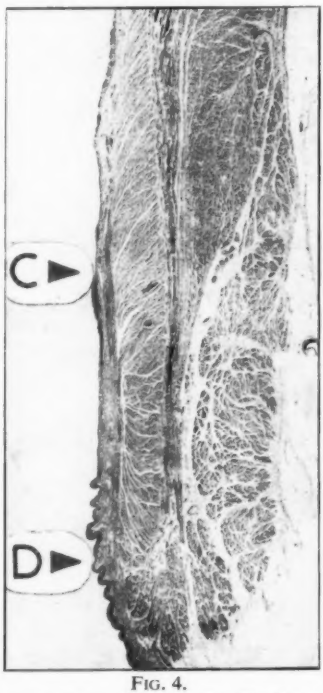

FIG. 4.

FIGS. 3 and 4.—Longitudinal section of anal canal taken at post-mortem. H.E. (× 4). The lower arrow in each (B, D) marks the level of the lower margin of the internal sphincter: the upper arrow (A, C) indicates the point of junction of squamous and columnar epithelium.

It is true, that as Wood Jones (Buchanan, 1949) points out, the skin of the lower half of the anal canal is not white but a colour contrasting between the bluish red of the rectal mucous membrane and the brownish pigment of the true perianal skin. One may point it out rather unconvincingly to a group of dressers as something which might pass for Hilton's line but it is not a line but a broad and ill-defined band of variable depth, and, further, it is seldom truly white. It has been stated that the line can best be seen in dark races (Kirk, 1947) but this assertion I have been unable to confirm. It is true that if one stains a section of the anal canal for melanin, the squamous epithelium can be shown to lose all its pigment a few millimetres below its point of junction with the mucous membrane (Figs. 7, 8, 9). It is possible to believe that this narrow ring of non-pigmented squamous epithelium may sometimes show up as a paler ring contrasting with the redder mucosa above and the pigmented squamous epithelium below. But even if this is so, it does not lie at the level of the intermuscular septum: the basal-cell layer at this level is always shown up quite clearly by the Masson stain

I naturally hesitated to question the existence of a white line, which was apparently sanctioned by the highest authority, and being disturbed in case I might be missing something which was clearly apparent to my colleagues I turned to the writings of our contemporary proctologists.

Gabriel (1948) in his great wisdom, I believe, makes no mention of either Hilton or his line, or of any line, white or otherwise. Lockhart-Mummery (1934) wrote of "a faintly marked line with a slight groove below it". Naunton Morgan (1936) places Hilton's line at the level of the anal intermuscular septum but goes on to say that "this much described line is only rarely seen".

I took some comfort from the self-confessed uncertainty of this evidence, and from that of others which is not dissimilar, and further in my search for the white line, I came across no photograph showing this distinctive anatomical feature and not even a convincing line drawing. Equally, when I have looked for it in the living patient I have found no hint of a white line but, at the most, an ill-defined pale-bluish ring of modified skin in the lower part of the anal canal which has in no way served as a useful anatomical landmark.

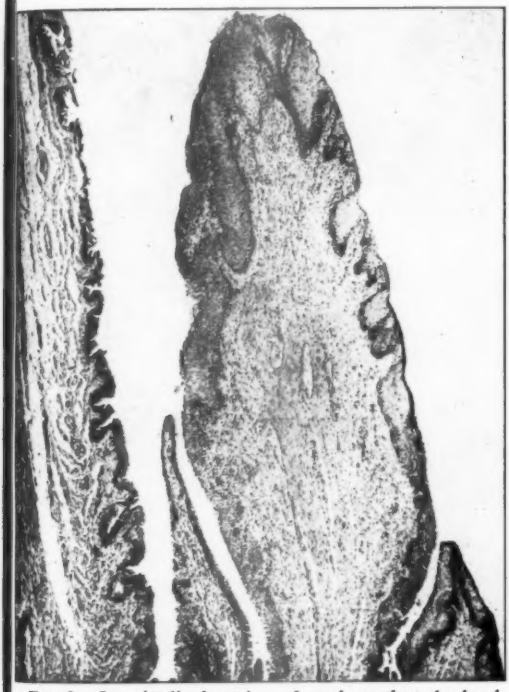

FIG. 5.—Longitudinal section of anal canal at the level of the valves to show the transition from squamous to columnar epithelium (× 32).

FIG. 6.—Engraving of the anatomy of the anal canal which illustrated Hilton's original text (f = a white line, e = internal sphincter, d = external sphincter). (Reproduced from the 1st edition (1863) entitled "On the Influence of Mechanical and Physiological Rest in the Treatment of Accidents and Surgical Diseases and the Diagnostic Value of Pain". By kind permission of the Royal College of Surgeons.)

CONCLUSIONS

(1) When Hilton in his 12th lecture spoke of the line of junction of the internal and external sphincters he rightly drew attention to an immensely valuable anatomical, and more especially a surgical, landmark. It is a level which, during life, we can be expected always to recognize and that with considerable precision.

(2) He was, however, wrong in believing this line to correspond exactly with the junction of mucous membrane and skin. It is always difficult to indicate exactly where this change takes place, but, as determined histologically, it is almost always at a higher level.

(3) The interval between the internal and external sphincters is not marked by a distinctive white line.

(4) The term Hilton's line is so ill-understood and so variously interpreted that it would be wise to discontinue its use altogether.

I wish to record my thanks to Dr. C. V. Harrison and to Mr. J-J. Brossy, F.R.C.S., for their generous help in the preparation of this communication.

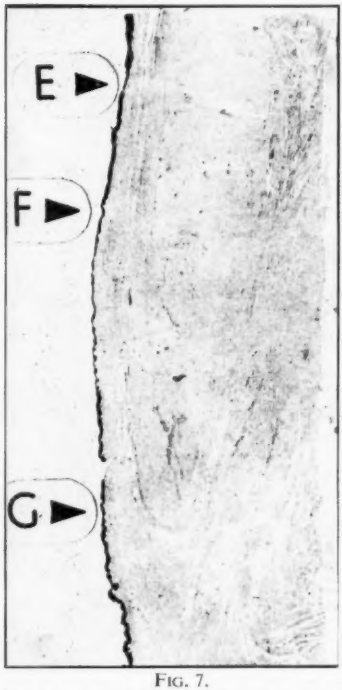

FIG. 7.

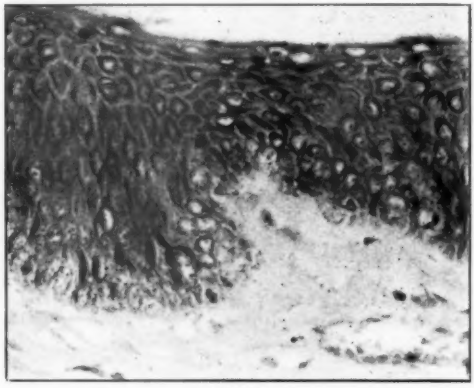

FIG. 8.

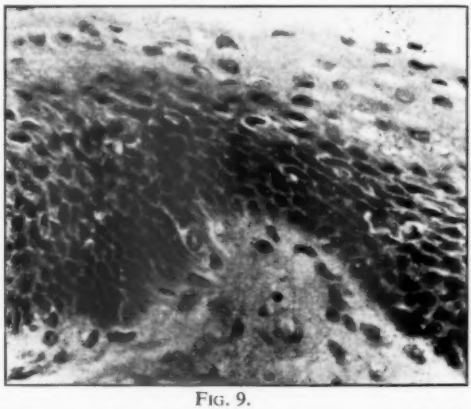

FIG. 9.

FIG. 7.—Longitudinal section of anal canal (× 4). E = junction of columnar and squamous epithelium. G = lower margin of internal sphincter. Stains for melanin show that the amount of pigment diminishes gradually between G and F, and between F and E it is completely absent.

FIG. 8.—Section (× 360) stained for melanin taken immediately *below* point F in Fig. 7, shows pigment in the basal layers.

FIG. 9.—Section (× 360) stained for melanin taken immediately *above* point F in Fig. 7, showing the complete absence of pigment granules.

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Section of Pædiatrics

President—C. T. POTTER, M.D., F.R.C.P.

[*November* 27, 1953]

DISCUSSION ON HIATUS HERNIA

Professor J. M. Smellie (The Children's Hospital, Birmingham): Infants suffering from what is now generally called "hiatus hernia" have as the outstanding clinical feature repeated vomiting usually dating from birth and the vomitus frequently contains some blood.

The term "hiatus hernia" implies protrusion of a portion of the stomach through the gastro-œsophageal opening in the diaphragm, so that the cardia comes to lie within the thoracic cavity. It is debatable, however, whether this anatomical malposition of the cardia is due primarily to an enlarged diaphragmatic opening with an upward retraction of the œsophagus or to a true œsophageal shortening as a consequence of a failure of normal growth and development. Until this controversial question has been settled it is preferable, at least in so far as infants are concerned, to use the term "partial thoracic stomach" thus avoiding any implication of precise ætiology (Carré *et al.*, 1952). The fact that there is a certain amount of mobility of the œsophagus in health must not be overlooked, as normal respiratory movements of the diaphragm do require the œsophagus to slide up and down for a short distance (Rios-Solans, 1953). When this defect is present at birth a congenital short œsophagus would appear to be the more reasonable explanation. This contrasts with a para-œsophageal hernia, which is clearly due to a pathological diaphragmatic opening, but only exceptionally is there any displacement of the cardia. When the lesion is first found in adult life it is more comparable to herniæ elsewhere and arises from an acquired incompetence of the hiatus consequent upon a loss of elastic tissue, increased intra-abdominal pressure or laxity of the diaphragm (Jones, 1952).

Be this as it may, the exciting and fundamental cause of the symptomatology of the disease is a failure of function of the gastro-œsophageal junction, permitting a reflux from the stomach into the œsophagus, with consequent emesis. This regurgitation of acid gastric contents may give rise to œsophagitis, ulceration, and eventual stricture. If, on the other hand, the cardiac sphincter mechanism is intact, whatever may be the position of the cardia the child will be asymptomatic and clinically well.

What mechanisms are responsible for the closure of the gastro-œsophageal junction is controversial. There is no true anatomical muscular sphincter at the lower end of the œsophagus, but the extent to which physiological closure is determined by anatomical structures, autonomic innervation or muscular contraction has yet to be elucidated. May it not be that different cases of "chalasia" of the cardia have different ætiological causes and that there is no one common factor operative in all instances? (Berenberg and Neuhauser, 1950; Wyllie and Field, 1946.)

Incidence.—The incidence of what I prefer to call partial thoracic stomach is by no means insignificant. For example, Birmingham Children's Hospital during the last several years has admitted some 15 to 20 infants each year suffering from this abnormality.

Symptomatology.—Repeated vomiting dating from birth or the early neo-natal period is the usual story.

Robert H., aged 9 days, with history of vomiting since birth, sometimes during, sometimes after, feeds. No blood present in vomits and feeds always taken well. X-ray shows a small loculus of thoracic stomach with marked gastro-œsophageal reflux (Fig. 1).

Initially the vomiting is closely related to feeds, is occasionally projectile and thus the picture may simulate pyloric stenosis. Within our experience the combination of partial thoracic stomach and hypertrophic pyloric stenosis has been observed in eight infants. It is interesting to speculate that the cardio-œsophageal reflux in such instances has been aggravated, if not caused, by the increased intra-abdominal pressure which is an inevitable complication of the pyloric obstruction. Blood-staining of the vomit is frequent and this has been observed occasionally within the first few days of life. Presumably it indicates some œsophageal ulceration and it is surprising that this can occur so soon after birth. After the neo-natal period the association between feeding and vomiting is not so close but change of position, crying and handling of the infant are more likely to promote vomiting. These infants fail to thrive but constipation is not nearly so constant as in pyloric stenosis. An appreciable number of infants with a partial thoracic stomach suffer little or even no inconvenience, therefore medical advice is not sought. For example, I have notes of one child who died of miliary tuberculosis at the age of 3 months. Routine post-mortem examination revealed a cardia 1 in. above the diaphragm, but there had not been any vomiting or feeding difficulties prior to the onset of the fatal illness. Occasionally confusion may arise with pyloric stenosis particularly when gastric peris-

talsis is visible, but blood-stained vomit is very rare in pyloric stenosis. In the older infant the picture may simulate a respiratory infection or even whooping-cough.

Prognosis.—Before an assessment of treatment can be intelligently measured some knowledge of the natural history of the malady is essential. This point is not infrequently overlooked and erroneous impressions of the benefit of treatment can thus be easily created. Our experience in Birmingham (Carré *et al.*, 1952) has demonstrated that in the large majority of these children the disease is benign and a symptomatic cure occurs in most.

The following case demonstrates a clinical cure though radiological examination shows a persistence of the regurgitation.

David E., admitted August 1950, aged 8 days. Vomiting started on first day of life, which was projectile and continued until admission. There had not been any blood in the vomit but this was observed within twenty-four hours of admission. X-ray shows fairly large loculus of stomach above the diaphragm. Ever since has been treated on expectant lines.

FIG. 1

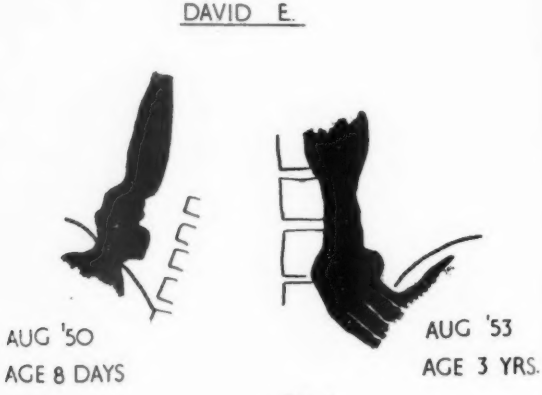

FIG. 2

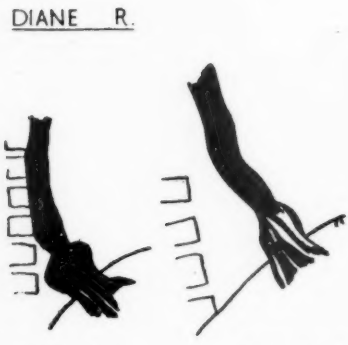

FIG. 3

FIG. 5

August 1953: Now 3 years old; very well, not vomiting.

X-ray: There is quite a considerable loculus of thoracic stomach with some gastro-œsophageal reflux, but no obstructive element (Fig. 2).

We are of the opinion that less than 10% of the cases diagnosed in infancy are likely to develop an œsophageal stricture.

The following case exemplifies clinical and radiological persistence of œsophageal regurgitation over a period of almost two years, yet there is no demonstrable œsophagitis or stricture.

Diane R., now 22 months old. Her vomiting has persisted and her response to nursing upright and with a mixed diet has not been very successful, though her nutrition is good (22 lb.). Vomiting did not start until she was a month old and vomits were large and forcible and blood was present. X-ray (March 1952) showed a large loculus of thoracic stomach and œsophageal irregularities suggesting œsophagitis.

X-ray: October 1953 (nineteen months later)—thoracic stomach persists and reflux present. There is no œsophageal stricture (Fig. 3).

A further detailed study and follow-up of some 50 cases in whom the diagnosis has been established during infancy is being undertaken by my colleagues, whose findings will be published in due course.

Treatment

Symptomatic and palliative measures.—Here two main principles have to be observed: (*a*) nursing the infant in as near as possible an erect posture day and night, and (*b*) thickening the feeds and introducing a semi-solid diet as soon as possible. It has not been found practicable to maintain a sitting position for these infants by means of pillows and in Birmingham we have devised a satisfactory, simple and cheap plaster chair which can be used both in a cot and in a pram (Hill, 1953) (Fig. 4). This chair has been used successfully both in hospital and in the home. It is very easy to make a new chair when the baby outgrows its original one.

FIG. 4 reproduced from *Lancet*, 1953, ii, 71, by permission of the Editor.

Treatment on these lines usually brings about a rapid abatement of symptoms and a satisfactory improvement in the nutritional state of the infant.

Cases where the disease has not been recognized until the age of 9 to 12 months may not respond so quickly but even so the results are promising and so far we have not had any reason to regret such expectant treatment.

Stephen K. First seen in March 1953, when 8 months old, with a history of repeated vomiting since birth with some blood in the vomit during the previous month. During succeeding seven months with nursing in erect posture he has gained 6 lb. in weight. Radiologically a partial thoracic stomach persists unchanged with some reflux but no signs of any œsophageal stricture (Fig. 5).

Craig (1953) has drawn attention to a flattening of the top of the head which he has observed in two infants who had been nursed upright for a period of four months. During the last several months we have carefully scrutinized our babies and have seen one infant—now 9 months old—whose skull does appear a little flattened. Whether this alteration in shape is due to treatment cannot yet be answered. There is, however, another point to which attention should be directed, namely, whether such a position may not have some deleterious influence upon the growth and development of the pelvis, important, in particular, so far as female infants are concerned.

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Dr. C. J. Hodson (University College Hospital, London) (Paper originally read at the International Congress of Radiology, Copenhagen, 1953): In adults a lax cardia with free reflux of gastric juice into the œsophagus and the sliding type of hiatus hernia can be demonstrated in about 5% of all barium meal examinations when the necessary technical procedure is carried out. There may be no symptoms.

In children, the condition usually presents between 5 and 10 years as œsophageal stenosis or ulceration and it is now believed that this represents the late stage of a process starting probably during the first year of life. Whether lax cardia and sliding hernia are as common at this stage as in adults is not known, because no series of examinations directed toward the elucidation of this point has yet been published.

In infants, of recent years, the attention of pædiatricians has been increasingly directed to the possibility of reflux œsophagitis as a cause of vomiting, and barium meal examinations have been carried out in many suspect cases. This contribution is based on the results of 100 consecutive examinations of this kind on infants under the age of 14 months. It must be emphasized that the cases were referred by experienced clinicians after other obvious causes of vomiting, such as pyloric stenosis or intestinal obstruction, had been excluded and are therefore a somewhat specialized group. Moreover, several were included in which the expectation of demonstrating an abnormality was thought to be small.

Table I shows the results of these examinations.

TABLE I.—ANALYSIS OF 100 BARIUM MEALS ON INFANTS

No abnormal findings		72
"Pathological" regurgitation		21
(*a*) Uncomplicated	10	
(*b*) *Œsophagitis*	11	
Other causes:		
Tracheo-œsophageal fistula		7
Duodenal atresia		
Pyloric stenosis, &c.		

Fig. 1 illustrates the age incidence of the cases examined.

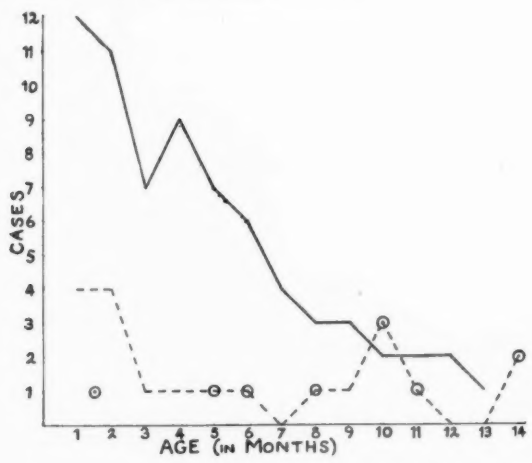

FIG. 1.—100 Barium Examinations on Vomiting Infants. ——No abnormal findings; - - - Excessive regurgitation; o Reflex œsophagitis.

Of the positive cases found, 2 settled down with conservative treatment; 9 underwent surgical operation, and of these 4 still require dilatation of the residual stenosis. These results amply demonstrate the reliability of the radiological method in the assessment of this condition.

The problem of radiological diagnosis is not so much the recognition of the advanced case, which is usually straightforward, but the separation of early abnormality from what can only be regarded as a wide range of normal variations. For simplicity the findings have been divided into four groups: (1) Abnormal œsophageal movement. (2) Excessive gastro-œsophageal reflux. (3) Changes in the wall of the œsophagus. (4) Herniation of the stomach. A wide hiatal opening has not been considered an intrinsic part of this picture as it is present in many cases of reflux where there are no secondary

effects. In the well-established case all four of these criteria are present in varying degrees of severity and it is the object of the barium examination to establish the presence and severity of each.

Œsophageal movements.—A study of the normal reveals a wide individual variation. In some cases there is vigorous effective peristalsis with rapid passage of a bolus down the œsophagus and straight into the stomach. In others there is a temporary hold-up just above the diaphragm by what looks like an efficient cardiac sphincter and more than one bolus may collect before emptying into the stomach occurs. In others again all movements are feeble and food tends to lie in the œsophagus until a series of peristaltic waves arises which pushes it on.

In the normal case also the junction between œsophagus and stomach is well defined, there being an efficient "sphincter" between the two, whatever its nature, which one can define even when relaxed. In the abnormal one with a wide hiatus it may be quite impossible to say where œsophagus ends and stomach begins, the whole being a continuous tube.

In the abnormal case the findings are again variable. There may be no abnormality seen until the stomach fills and reflux commences. Or, there may be very little movement at all and the œsophagus may act as an inert tube up and down which regurgitated fluid passes at each respiration in the supine position, what motive force there is being due mainly to the act of swallowing or to gravity when the patient is erect. In others the movements are more vigorous but unco-ordinated and die away in the lower third of the œsophagus. In 5 of our cases there was well-defined localized spasm in the lower œsophagus which partly relaxed to allow fluid to pass, and in 2 there was a classical incisura opposite an ulcer crater, such as is seen in peptic ulceration in the stomach or duodenum. In the advanced case, of course, the œsophagus is dilated proximal to the stenosis.

Regurgitation.—In the normal infant when the stomach is full and the child lies in the supine position reflux takes place at irregular intervals without any warning, and is usually seen two or three times during the period of examination *which is ten to fifteen minutes.* In some cases it occurs more frequently when a large amount of air is present in the stomach, and ceases when the air has been expressed. Crying also tends to increase the frequency of reflux.

In this series a pathological state of affairs was considered to be present when reflux took place freely at every deep inspiratory phase of respiration. 21 of the cases showed this state of affairs, some with associated structural change, some without. It was found unnecessary to adopt the Trendelenburg position; to hold the child upside down; or to exert pressure on the stomach to demonstrate this finding. All that was needed was a full stomach.

Changes in the œsophageal wall.—These were usually striking and accompanied by the conditions just described. In their simplest form they consisted of rigidity of the œsophageal wall with localized

FIG. 2.—Constant "toothlike" projection from œsophageal wall believed to indicate œsophagitis.

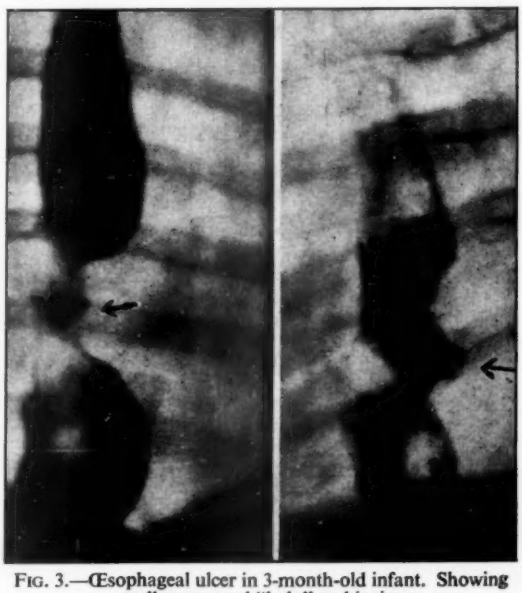

FIG. 3.—Œsophageal ulcer in 3-month-old infant. Showing swollen mucosal "halo" and incisura.

impairment of peristalsis. This rigidity was sometimes associated with constant sharply-angled tooth-like projections from the wall and these are now believed to be a reliable sign of œsophagitis (Fig. 2). In 3 cases there were obvious ulcer craters, such as we find in peptic ulceration elsewhere (Fig. 3), and in 2 other cases there were probably ulcer craters. In at least 2 cases there was evidence of the œsophagus being adherent to surrounding structures which suggested the adhesions that are sometimes met with at operation.

Finally, in the more advanced cases there was marked stenosis and shortening of the œsophagus such as is well known in older children, and twice it was shown that this advanced state developed in a period of only three or four months. The length of this stenosis is usually only 1 to 2 cm. but may be as long as 4 or 5 cm., and in 2 cases two narrowed lengths were found and proved at operation.

Herniation of the stomach is usually easy to demonstrate in the advanced cases providing the stenosis allows the barium fluid to pass through for its delineation. It is not always easy to demonstrate in the early stages when there is no well-defined line of demarcation between œsophagus and stomach. I have found a study of the peristaltic wave and the folds of the endothelium to be unreliable guides in this problem and I now usually settle the point by carrying out bi-manual compression when the stomach is full. This manœuvre readily produces herniation when it is of the intermittent type and exaggerates it when it is permanent.

In the 11 cases labelled "œsophagitis" these four criteria (free reflux, abnormality of œsophageal movement, structural changes in the œsophagus, and hernia) were all satisfied. Where possible they have been confirmed by œsophagoscopy, operation, or by follow-up examinations.

Technique.—Because the cause of vomiting is unknown when the baby is first examined the method used must be capable of excluding disease at least as far as the small intestine and it must also be able to establish the four criteria mentioned. Radiographs of the chest and abdomen are first taken to exclude gross changes and intestinal obstruction. The child is then bottle or spoon fed with a preparation containing barium sulphate, sugar and dried milk, which is usually greatly appreciated, and as a full stomach is essential for adequate demonstration, this is an important point.

The œsophageal movements are first observed fluoroscopically in the supine position. The child is then turned on its right side while feeding continues and kept so until the pylorus opens, when the duodenum is examined. Pyloric stenosis can thus be excluded. This is an important matter as there is a distinct association between pyloric stenosis and gastric reflux. If air collects in the stomach during feeding it is expelled in the erect position. When the stomach is full the child is again placed on its back and the amount of regurgitation noticed. Bi-manual pressure is then applied to demonstrate the presence or absence of a sliding hernia. Finally an eighteen-hour film is taken to exclude such things as malrotation of the intestine. The whole process takes ten to fifteen minutes and no ill-effects have been noticed.

SUMMARY

An analysis of 100 consecutive cases, referred for barium examination because of unexplained vomiting before the age of 18 months, showed that 11% had evidence of œsophagitis caused by an abnormal degree of regurgitation of gastric juices. Later examination showed, as has been the experience elsewhere, that this can lead rapidly to the severe stenosis for which the present remedies are both drastic and rather unsatisfactory, indicating the necessity for the earliest possible recognition.

A cine-radiographic film was shown to illustrate the points raised.

Mr. David Waterston (Hospital for Sick Children, Great Ormond Street): There has been some controversy as to whether hiatus hernia is a congenital disease. Fig. 1 is a radiograph of a baby who was admitted when 1 day old with symptoms suggestive of a tracheo-œsophageal fistula; it shows well the typical appearance of a hiatus hernia with an intrathoracic loculus of stomach. On numerous occasions we have been able to demonstrate this lesion in the newborn, both radiologically and at operation.

There seems to be no doubt therefore that the lesion may be present at birth and probably before; may it not be possible that some of the complications of hiatus hernia may occur *in utero*? If we believe that the sequence of events in some cases of hiatus hernia is: reflux of hydrochloric acid—œsophagitis—ulceration of lower œsophagus—stricture formation, why should not this process occur before the child is born? All the requisites are there: the child is in a head-down position making reflux easy; there is often a very high concentration of hydrochloric acid in the stomach in the last few weeks of gestation. Why should not stricture formation also occur? This might account for the well-described cases of web-like obstruction in the œsophagus in the newborn.

All the strictures of the œsophagus in children that I have seen (apart from those caused by corrosive fluids or trauma) have been in cases with a hiatus hernia; the pouch of stomach above the diaphragm is not always easy to demonstrate radiologically, but can always be seen at operation.

Fig. 2 shows the types and complications of hiatus hernia that may be seen in children. The stomach in Type C may undergo volvulus either above *or below* the diaphragm.

Indications for Surgical Treatment

Since the majority of these children lose their symptoms when they start walking upright, the only indication for surgical treatment is the presence or the threat of complications. Œsophageal ulceration which progresses to fibrous stricture alters the prognosis so profoundly that every effort should be made to prevent this complication. The assessment of the severity of the œsophagitis and the recognition of a stricture once it has formed can only be made by œsophagoscopy. This examination should be carried out in all children of whatever age, if œsophagitis is suspected.

Hæmatemesis by itself is no indication of the severity of the ulceration, and the child on a fluid diet who appears to be getting better because he is vomiting less frequently, may owe his symptomatic improvement to the formation of a stricture in the œsophagus.

FIG. 1.—Hiatus hernia. Aged 3 days.

Anatomy

There are several interesting anatomical details seen at operation which may have a bearing on the ætiology of the condition.

(1) *Length of œsophagus.*—In all the cases operated upon it has been possible without difficulty to replace the cardia below the diaphragm; that is to say the œsophagus did not appear to be truly "short".

(2) *Vagus nerves.*—In many cases the vagus nerves appeared "slack" and only took up their normal tightness after the cardia had been stretched to below the diaphragm.

(3) *Infracardiac bursa.*—In over half the cases operated upon a serous-lined bursa was opened, upon which the intrathoracic loculus of stomach glided. This bursa lies upon the right crus of the diaphragm; it is separate from peritoneal and pleural cavities, and is described anatomically as a persistence of the cephalic part of the pneumato-enteric recess (Gray, 1949).

(4) *Absence of peritoneal sac.*—Only in cases shown in Fig. 2C is a sac of peritoneum drawn up into the chest with the stomach. In the more common smaller type Fig 2B there is no peritoneal sac surrounding the hernia.

(5) *Size of hiatus.*—In all cases operated upon the hiatus seemed larger than normal. Following the replacement of the stomach the hiatus was narrowed posteriorly with three or four silk stitches.

Associated anomalies.—Other congenital anomalies are rare. 4 cases had congenital pyloric stenosis; 5 cases had abnormalities of the glottis or trachea producing stridor.

SURGICAL TREATMENT

The surgical repair of the hiatus and the replacement and fixation of the stomach below the diaphragm are difficult; the technique of operation does not come within the scope of this paper. The results of treatment in this condition are difficult to assess. As the symptoms tend to disappear without treatment and long remissions from vomiting are common, a large number of cases and a very long term follow-up is necessary before any conclusions about treatment can be made. I therefore look on these figures and results as only an interim report. The follow-up of these cases is still continuing.

Total cases of hiatus hernia in children	132
Cases treated surgically	62

All these cases have been operated on by Professor R. S. Pilcher or myself.

Cases with stricture (i.e. about ⅓ of operated) ..	20
Recurrences after operation	6

3 of these were in the first 15 cases after which the technique of repair was modified.
5 of these recurrences have been re-repaired.
One has recurred radiologically but has no symptoms.

FOLLOW-UP

11 cases are too recent for adequate follow-up. 51 cases have been followed for over six months. Of these—32 may be classed as good results. They have no vomiting or pain and are gaining weight. 11 have fair results, i.e. the parents are pleased, but they still vomit occasionally and although they have no hæmatemesis 5 still show a mild œsophagitis on œsophagoscopy. Some of these cases may get worse with a longer follow-up. 8 cases still have symptoms, and have not been improved. They have recurrent attacks of severe œsophagitis with bleeding; most of them have pain.

I suspect that most of this last group fall into the group described by Barrett (1950) and Allison (1951, 1953) who have extensions and islets of gastric mucosa extending up the œsophageal tube. Most of them will probably require more radical surgery.

There have been no deaths in this series, but it should be stressed that the operative repair of these hernias is a major procedure and is bound to carry the risk of any thoracotomy. In our experience thoracotomy for any condition is better tolerated and has fewer post-operative complications in the group under 2 years than over 2 years of age.

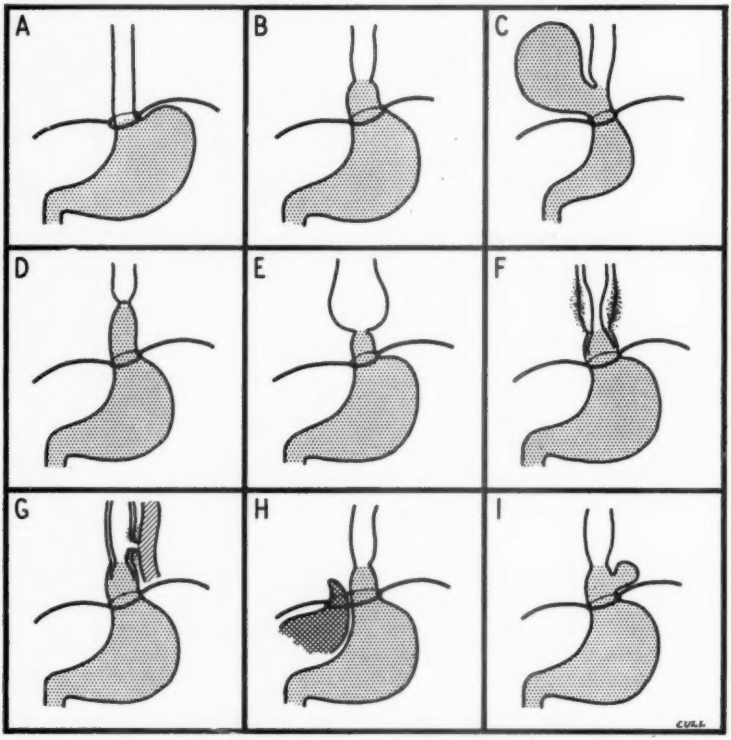

FIG. 2.—Varieties and complications of hiatus hernia seen in childhood: A, Normal. B, Hiatus hernia. C, Hiatus hernia with large intrathoracic loculus. D, "Congenital web" in œsophagus. E, "Congenital stricture" in œsophagus. F, Hiatus hernia with œsophagitis and peri-œsophagitis. G, Hiatus hernia with œsophageal ulceration involving aorta. H, Hiatus hernia with prolapse of liver through hiatus. I, Hiatus hernia with diverticulum of intrathoracic loculus.

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Dr. R. Astley (The Children's Hospital, Birmingham): I feel that Dr. Hodson's diagnostic criteria must be too strict if he finds that each of his abnormal children shows reflux with every breath taken. We have had a considerable number of children where it is much more difficult than this (or even impossible) to demonstrate their gastro-œsophageal reflux, yet they have had a definite loculus of stomach above the diaphragm.

From a radiological viewpoint I feel that we have to consider three groups of abnormalities at the lower end of the œsophagus:

(1) A loculus of thoracic stomach of greater calibre than the œsophagus.

(2) A tubular thoracic stomach, where the gullet appears very much the same calibre down to the diaphragm but its lower portion is gastric, as evidenced by mucosal pattern studies.

(3) Gastro-œsophageal incompetence with minimal detectable anatomical abnormality.

These are not hard and fast groups as transitional forms occur and the appearances may sometimes vary somewhat from examination to examination.

In the second group, the tubular thoracic stomach, come many of the cases that have been reported as cardio-œsophageal relaxation or "chalasia". Indeed the illustrations to some of these alleged cases of "chalasia" show that there is really gastric mucosa above the diaphragm although the authors have not realized it.

The third group forms the real problem for the radiologist. There are undoubtedly a number of babies and children in whom gastro-œsophageal incompetence is present as evidenced by the clinical story, which may be exactly the same as that in the other two groups. However, the incompetence may or *may not* be shown by gastro-œsophageal reflux during screening. Moreover, there is no definite loculus of stomach above the diaphragm, although often there is a suspicion of anatomical abnormality. The stomach may appear a little "peaked up" towards the hiatus and there may be the suspicion of gastric mucosal folds just above the diaphragm, but the abnormality is indefinite.

Undoubtedly it is sometimes a failure or inadequacy of the method that is responsible for such diagnostic doubt. If, at the time of examination, reflux is slight or absent, a loculus of thoracic stomach cannot always be fully distended with barium and its true nature revealed. This is shown by the failure to detect for certain a thoracic stomach at one examination but its recognition at a later date when reflux is more evident. By rapid serial pictures, taken as for angiocardiography, in some individuals with a partial thoracic stomach it can be shown how a normal appearance of the barium may occur at some moments and an abnormal picture at others, according to the exact degree of filling with barium.

Thus the minimal anatomical changes in some of my third group can sometimes by repeated examinations be proved to indicate a real loculus of stomach above the diaphragm. In others, their true significance is never discovered. We have had approximately 20 such indeterminate cases, as compared with just over 150 who had a definite partial thoracic stomach.

The moral of this is that if there is a suggestive clinical history a single negative barium swallow should not be accepted; X-ray examination should be repeated several times before the gastro-œsophageal junction is considered to be normal.

With regard to prognosis, there seems to be no co-relationship between this and the degree of reflux as observed during fluoroscopy. As Professor Smellie has said, about three-quarters of our cases in Birmingham have had a relatively benign course, with symptomatic cure during the first two years of life. However, the presence of gross reflux at the onset does not exclude a child from such rapid improvement, nor does minimal reflux exclude a more prolonged course. The development of an œsophageal stricture does, of course, mean a worse prognosis but even here it may be pointed out that some of these can disappear spontaneously.

Dr. Hugh Jolly questioned Professor Smellie's statement that hæmatemesis was rare in pyloric stenosis. During the past year in Plymouth he had seen 3 such cases out of a total of 22 and he thought that this might have been due in some instances to a delay in treatment, with consequent severe gastritis, resulting from the greater distances involved in Devon and Cornwall.

Dr. Jolly also discussed the question of œsophageal stricture as an indication for immediate operation. A stricture seen on barium swallow was not necessarily permanent and might be the result of spasm and œdema rather than fibrosis. For this reason he felt that a period of medical treatment should always be tried in these cases. No one denied the ultimate need for surgery where a stricture existed but this preliminary medical treatment gave the opportunity to improve the child's general health and possibly to alter the type of repair if the stricture disappeared. He showed X-rays in which an apparent stricture had completely disappeared in six months of medical treatment while the child's general condition had been transformed.

Dr. Hodson, in reply to Dr. Astley, said that unless strict criteria were adopted it became very difficult to say where normal variation ended and disease began and that as a severe operation might depend on the results it behoved one to be strict. If small hernias might thus be dismissed at least the all-important œsophagitic changes were identified, and it was on these that the seriousness of the lesion rested.

Professor Smellie, replying to Dr. Jolly, reiterated that within his experience hæmatemesis was rare in pyloric stenosis but did agree that bleeding could occur, particularly in severe cases, if there had been delay in recognition of that disease. He also concurred with Dr. Jolly's opinion that spasm and œdema of the lower œsophagus could produce a radiological picture simulating that of an organic stricture and operative procedures should not be advised before the results of expectant medical treatment had been ascertained.

[*January* 22, 1954]

Chondro-ectodermal Dysplasia (Ellis-van-Creveld Syndrome). Colobomata of Iris.—MALCOLM MACGREGOR, M.D.

FIG. 1

D. G., male, aged 7.

Family record.—Only child. Father Irish—colobomata of iris. Mother English, healthy. Mother has re-married and has since had one normal child.

Past illnesses.—Measles. Amputation of 6th digits from each hand aged 3. Broke leg after minor trauma last year. Average at school.

General characteristics (Fig. 1).—Weight 43 lb. Height 42 in. (average 47 in.). Symphysis to vertex 26½ in. (average 25 in.). Symphysis to soles 16½ in. (average 22 in.). Intelligent and lively. Dwarfed. Short thick limbs with lateral bowing of proximal segments. Knock knees. Mild pigeon chest; Harrison's sulcus. Heart sounds normal. Scars on outside of hands from amputated digits. Restriction of movement at interphalangeal joints. Bilateral incomplete colobomata of iris. Teeth: some incisors absent; others of abnormal shape. All teeth carious and unduly subject to decay. Finger-nails small and dystrophic. Toe-nails thickened and abnormal.

X-rays show that dwarfism is solely due to shortening of lower extremities. Spine, skull and chest normal. Carpus normal. Upper limbs: outward bend of humeri. Radius and ulna short. Squat metacarpals and proximal phalanges with epiphysis embedded in deep cups. Fifth metacarpals show lateral prominence at site of sixth digit. Lower limbs: all bones short. Through cortex of these bones pseudo-fractures (Looser's zones) are visible. Irregular condensation and rarefaction at metaphyses of tibiæ. Upper epiphysis of tibiæ flattened laterally causing valgus deformity. Metatarsals and phalanges resemble condition in hands. Teeth: many front teeth misplaced, unerupted or poorly developed.

Other investigations.—Serum calcium 9·5 mg.%. Serum phosphorus 4·1 mg.%.

Chronic Myeloid Leukæmia.—A. D. M. JACKSON, M.D., M.R.C.P., D.C.H. (for R. H. DOBBS, M.D., F.R.C.P.).

K. T., male, born August 17, 1941.

History.—Admitted to the London Hospital in March 1952 (aged 10½ years) complaining of anorexia, lassitude and loss of weight of six weeks' duration.

On examination.—There was pallor of the skin and mucous membranes and enormous enlargement of the spleen (Fig. 1). There was no enlargement of the liver or lymph glands and no evidence of a hæmorrhagic state.

FIG. 1.—The shaded area shows the size of the spleen at the time of the diagnosis.

Investigations.—Hb 49%. R.B.C. 2,600,000 per c.mm. Colour-index 0·9. W.B.C. 250,000 per c.mm. (polymorphs 47·5%, lymphocytes 6·5%, eosinophils 1%, metamyelocytes 7·5%, neutrophil myelocytes 15·5%, premyelocytes 10%, myeloblasts 12%). Platelets normal. Urine normal. X-rays of long bones normal.

Progress.—Deep X-ray therapy was given to the spleen in a total skin dosage of 350 r spread over three weeks and the patient was discharged from hospital symptom-free and gaining weight. The total white cell count fell rapidly, reaching a normal level with only a small percentage of primitive cells, about two weeks after the cessation of treatment. The changes in the blood count during the progress of the disease are shown in Fig. 2. At no time did the platelet count fall below normal. There was considerable reduction in the size of the spleen although it remained enlarged.

For the next eighteen months the patient remained well and lived a normal life, attending school and playing games, but one year after the initial diagnosis the white cell count began to rise with an increasing

percentage of primitive cells and the spleen became more enlarged. In December 1953 the boy was readmitted to hospital with a white cell count of 288,000 per c.mm. and a further course of deep X-ray therapy was started.

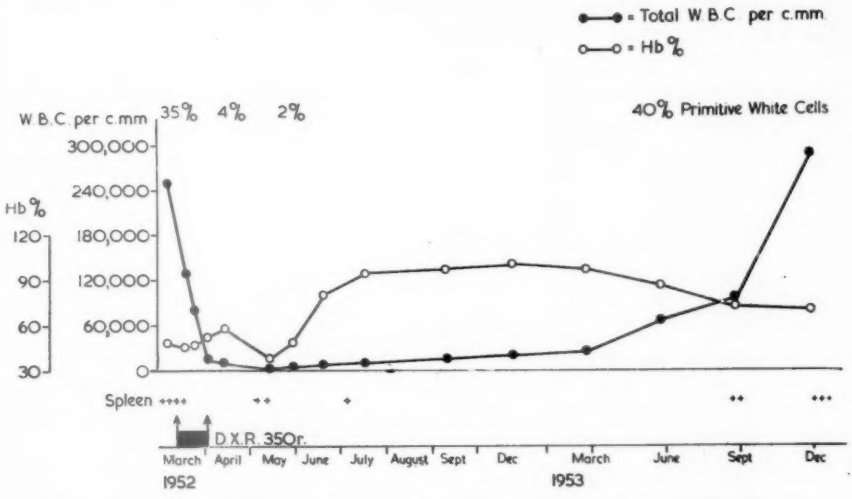

FIG. 2.—K. T., aged 12½ years. Chronic myeloid leukæmia. Showing the course of the disease.

Comment.—The rarity of chronic myeloid leukæmia in children is emphasized by two recent reports. Cooke (1953) collected 294 cases of leukæmia in children over a period of twenty years and of these only 10 were of the chronic myeloid type. Gauld *et al.* (1953), analysing all the recorded cases of leukæmia between 1938 and 1951 in hospitals in Aberdeen and Edinburgh, including the Hospitals for Sick Children, found 122 cases of leukæmia in children under 15 years of age. This figure included only 2 cases of chronic myeloid leukæmia.

The present case illustrates the course of the disease when treated with radiotherapy, the patient having so far survived for two years from the onset of the disease to live a normal life with only two brief periods in hospital.

POSTSCRIPT.—At the beginning of February 1954 (i.e., two years from the onset of the disease) the patient was again discharged from hospital after a four-week course of radiotherapy (total skin dosage 1,400 r). There were no symptoms, the Hb was 72% and the W.B.C. 17,400 per c.mm. (17% primitive cells).

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Osteodystrophy Due to Congenital Deficiency of Phosphatase.—J. LUDER, M.D., M.R.C.P. (for B. E. SCHLESINGER, M.D., F.R.C.P.).

C. A., female, aged 15 months.

History.—Normal progress until 2 months when she began to vomit and became reluctant to take feeds. The symptoms persisted and her weight gain was unsatisfactory. At 7 months she was admitted to hospital having become slowly worse in the previous three weeks, and found to be wasted, with a temperature of 100° F., and a bulging fontanelle. The C.S.F. was normal and she was transferred to the Hospital for Sick Children, Great Ormond Street.

Clinical findings.—A wasted dehydrated baby, weighing 10 lb. 8 oz. The skull circumference was 16½ in., frontal bossing was present and the anterior fontanelle was large and bulging. No signs of meningeal irritation were present and the discs were slightly pale, without papilloedema.

There was enlargement of all the costo-chondral junctions on both sides, forming a rickety rosary, enlargement of the lower ends of the radius and ulna on both sides and slight bowing of the tibiæ.

The skin showed mottled brown pigmentation of the hands and face and the scleræ were blue. Other systems normal.

Progress and investigations.—Several days after admission a series of left-sided convulsions occurred during two days. Subdural taps were negative but air ventriculography revealed displacement of the

third ventricle and septum pellucidum to the left. Craniotomy was done and a thin right subdural hæmatoma was evacuated.

Skull X-rays taken for the ventriculogram showed an extraordinary degree of irregular decalcification of the bones of the vault with wide separation of the sutures. X-rays of the rest of the bony skeleton showed a similar condition widespread throughout all the skeleton and most marked in the metaphyses of the long bones (Fig. 1). Early periosteal reaction was present in some long bones.

FIG. 1.—Showing severe decalcification of metaphyses and periosteal reaction.

FIG. 2.—Showing improvement in calcification and periosteal new bone formation.

Blood chemistry.—The main findings were as follows: Serum calcium 14·8 mg.%. Serum phosphorus, 3·8 mg.%. Serum alkaline phosphatase 9 K. A. units. Blood urea 70 mg.%. Serum proteins normal.

Urine: A few pus cells and a moderate growth of *B. coli*. No amino-aciduria. Sulkowitch reaction normal. W.R. and Kahn reactions negative.

Bone biopsies (Dr. R. Pugh).—Skull, upper end of tibia: A mixture of lamellar and woven bone. No abnormal amount of osteoid but bone stains unevenly and there is an unusual feature in the presence of imperfectly calcified connective tissue surrounding the bone trabeculæ. Osteoblasts were plentiful, and osteoclasts scanty.

Calcium and phosphorus balance.—Both calcium and phosphorus balances were positive.

Treatment was begun with vitamin D, starting with 10,000 units daily, and increasing to 50,000 units daily, with a total dose of 3,630,000 units in three and a half months.

Bone X-rays showed much deposition of calcium at the hitherto uncalcified bone ends and there was much more marked periosteal reaction and calcification, leaving a clear zone underneath it (Fig. 2). The diaphyses became more homogeneous but the degree of calcification remained poor. The skull looked more normal and the fontanelle gradually closed.

The serum calcium fell slowly to normal during treatment, but then rose again to 11·5 mg.%.

The serum phosphorus remained normal for several months but then rose with the serium calcium to 6·8 mg.%. For these reasons, vit. D therapy was stopped.

The serum alkaline phosphatase showed a progressive fall to a figure of 3·8 K. A. units before discharge.

The blood urea remained raised at 50–80 mg.%.

The general condition of the child showed little change. There was a weight gain of 6 lb. in seven months but vomiting persisted. At fifteen months she could sit up but not stand and there were no words.

Tests for presence of phosphatase in bone.—(1) *Histochemical* (Dr. R. Pugh): Biopsy specimen from tibial metaphysis. No phosphatase demonstrated.

(2) *Chemical estimation* (Dr. J. W. Wilkinson): Tibial metaphysis. Phosphatase concentration of 150 nitro-phenol-phosphate units in bone cortex and periosteum. This was 29% of a normal control.

Discussion.—The possibility of widespread infiltration of the bone-marrow with malignant deposits such as secondary neuroblastoma, multiple myelomatosis, or leukæmia was ruled out by the bone biopsies. Hyperparathyroidism seemed a likely diagnosis at first but the bone biopsies did not support this, and the positive calcium and phosphorus balances put it out of court.

Conditions such as Ollier's disease and metaphyseal dysostosis were ruled out by the abnormal blood chemical findings.

Congenital hypophosphatasia was suggested by a report by Rathbun (1948) of a case showing a similar widespread osteodystrophy and a persistently low serum alkaline phosphatase. Blood calcium and urea were both raised. Sobel *et al.* (1953) published a review of these cases and added a further case, the common features being skeletal deformities resembling rickets, poor bone calcification, loss of milk teeth, and persistently low serum alkaline phosphatase. The possibility of poisoning by metals was ruled out, and the suggestion was made that this may be a genetically determined and possibly inherited characteristic.

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Cherubism.—A. WHITE FRANKLIN, M.B., F.R.C.P.

R. M., male, born 10.6.48, was brought to Osterhills, St. Albans, because of loss of permanent teeth and swelling of the face. He was born at term, weighing 7 lb. 13 oz., and was bottle fed. At 5 weeks he was admitted to hospital for ten days with pneumonia. His infancy had otherwise been uneventful except for mild measles and chickenpox. Anxiety began when he lost some milk teeth at the age of 3 years and became alarm when at the age of 5 years he shed his two recently erupted lower central incisors. The mother cannot remember first noticing the swelling of the face, but it was certainly present at 3 years.

Examined at 5 years (10.6.53) (Figs. 1 and 2): A well-developed boy, weight 39 lb., height 42½ in. The lower part of the face from the level of the zygomas to the chin is swollen and the eyes are full and tilted a little heavenward. The swollen alveolar ridges have produced a V-shaped hard palate, so

FIG. 1

FIG. 2

that the space in the mouth is limited. Small nodules, firm and painless, can be felt over the mandible, and sub-mandibular lymph nodes are enlarged. The rest of the physical examination is negative.

Family history.—Mother, father and sister of 10 years appear normal. A baby sister born September 1953 has a full lower face, but normal jaws in the X-ray.

Investigations.—X-rays of the jaws (Fig. 3) show gross enlargement of both jaws with diffuse cystic areas of rarefaction. Mr. J. D. Cambrook (June 1953) reported on the teeth:

Temporary erupted $\frac{CBA|ABCD}{DCB\ |\ BCD}$ Permanent unerupted $\frac{654321|123456}{6\ 43\ |\ 34\ 6}$

X-rays of skull and the rest of the skeleton normal. W.R. and Kahn negative. Hb 88%. W.B.C. 8 600 per c.mm. Serum calcium: 10·7 mg.%. Serum inorganic phosphorus: 5·2 mg.%. Serum alkaline phosphatase: 22 K.A. units.

Discussion.—This boy has the same appearance and radiological changes in the jaws as have been described by W. A. Jones of Kingston, Ontario (1933) and named cherubism. Jones first described 3 and later (1938; Jones *et al.*, 1950) 4 out of the 5 siblings in a family of Russian Jews. He regards it as a familial disease of the jaws, the onset at the end of the second or early in the third year. The lower jaws in his cases were operated on by Dr. J. Gerrie with improvement in appearance. Over the years the upper jaws have regressed and Jones (1953) believes that the condition is to some extent self-limiting.

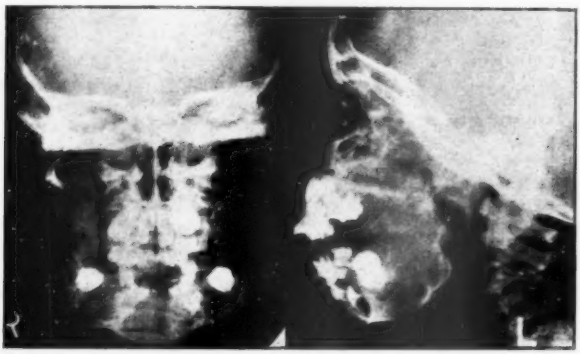

FIG. 3.—Antero-posterior and lateral X-ray of jaws showing gross enlargement and diffuse cystic areas.

In his second paper Jones (1938) quotes a case, referred to him by Dr. Thomas of Savannah, of a boy of 11 years, the only sufferer among his own siblings from the condition, traced back through five generations. In 1951 Caffey and Williams reported 5 cases in two families, one white and one coloured, although only one case in each showed the fully developed condition when examined, a boy of 5½ and one of 26 months. In a girl of 14 and a man of 33 the upper jaws including the teeth were apparently normal. Sir Thomas Fairbank (1952) refers to a possible case seen by Professor S. L. Baker in a boy of 8¾ years, who had in addition cysts in both humeri and both femora.

The clinical features are marked symmetrical fullness of the cheeks and jaws, slight upward tilting of the eyes, revealing white sclera below, and noticeable submaxillary swelling from enlarged lymph nodes. The jaws feel hard and are painless. The teeth are irregularly placed with many missing. The alveolar ridges are swollen, producing a narrow V-shaped hard palate. The intelligence, the rest of the skeleton and the blood chemistry are all normal.

X-rays of the jaws show gross enlargement with numerous sharply defined multilocular areas of decreased density. In many parts only a few opaque septa are seen. The teeth are irregular and as age advances many permanent teeth are missing.

Histology of the enlarged lymph nodes (Jones, 1933) shows lymph follicles well preserved, but thickened capsule, diffuse fibrosis and endothelial proliferation.

The bone at operation shows a loss of periosteum with fatty fibrous tissue escaping into and mingling with the tissues of the cheek. The bone is thick and hard, but the fibrous fatty tissue can be easily scraped away. The bone sections, described in detail in Caffey and Williams' paper (1951), show a cellular structure with many multi-nucleated giant cells of osteoclastic type.

There is in all cases except the present one, a positive family history. Although the first cases occurred in a Jewish family, cherubism has been found in a negro family and in the general white population in U.S.A. and now in England. The patients are normal clinically and radiologically until about 18 months to 2 years of age, with progressive swelling during childhood and regression during adolescence to a condition that would pass as normal in adult life, so that operation seems unnecessary. The lower jaw is affected more than the upper. The main disability is loss of teeth in the lower jaw.

Cherubism is probably a genetically determined defect of bone development localized to the jaws.

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Hirschsprung's Disease in a Man Aged 23.—J. J. KEMPTON, M.D., M.R.C.P.

The relative rarity, at present, of cases of this condition surviving to adult life, together with the fact that history is available dating from early infancy, has seemed to justify the placing on record of this case.

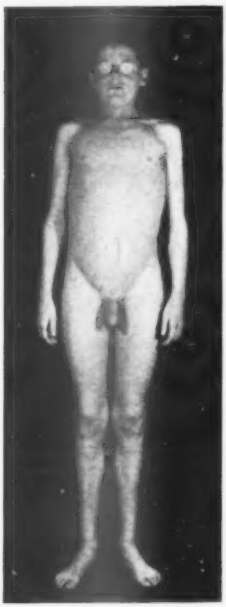
CASE REPORT

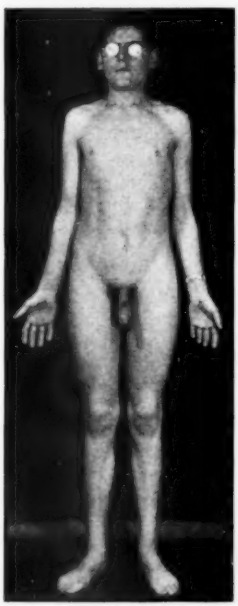
R. S., male, aged 23. Birth was normal and there is no relevant family history. He presented in the first weeks of life with constipation and episodes of distension and vomiting relieved in part by frequent enemas. During his first three years he was admitted on numerous occasions to King's College Hospital under the care of, first Sir Frederick Still, and later Dr. Wilfrid Sheldon, with recurrent attacks of obstruction. Colonic distension and peristalsis were first observed at the age of 4 months, and the diagnosis of Hirschsprung's disease was made at that time. The signs became more apparent later (Fig. 1) and a barium enema showed a grossly dilated colon.

FIG. 1.—R. S., aged 1 year 5 months, showing colonic distension and peristalsis. Photograph from King's College Hospital notes.

At 2½ sympathectomy was performed by Mr. (now Sir Cecil) Wakeley; the sympathetic chain and ganglia in the region of the superior mesenteric artery were removed, and the aorta near the origin of the inferior mesenteric artery was stripped of periarterial plexus and swabbed with absolute alcohol. This produced no apparent improvement, but after the age of 4 episodes of obstruction gradually became less frequent and less severe, though there were many further admissions to various hospitals in the London area.

At the age of 11, after one such admission to hospital, he was sent to a Hospital School on the Berkshire Downs for a period of convalescence. He remained there for a prolonged period, continuing





FIGS. 2 and 3.—Aged 20 showing some abdominal distension and absence of pubic hair. Eight months later (Fig. 3) considerable development has occurred after a period of washouts. Photographs reproduced with permission of English Universities Press, Ltd., from Le Marquand, H. S., and Tozer, F. H. W. (1954) Endocrine Disorders in Childhood and Adolescence. 2nd ed. London.

to have occasional enemas and with no further episodes of acute obstruction, and first came under observation at the Royal Berkshire Hospital at the age of 20. By this time he had ceased to be regarded as a patient, and worked in the garden, though occasional enemas were still given. He presented at this time all the characteristic appearances of the condition. There was flaring of the rib margin and abdominal distension (Fig. 2), and very large masses of soft fæces were palpable in his abdomen. He was short (62 in.) and of poor colour and muscular development. There was no pubic or axillary hair, though genital development was probably normal. His voice was high-pitched and radiological examination of his epiphyses gave a bone age of 14. There was a mild hypochromic anæmia (Hb 60%). After admission to hospital and bowel washouts, which removed immense quantities of fæces, films were obtained showing the typical appearance of Hirschsprung's disease (Fig. 4).

FIG. 4.—X-ray showing inactive terminal segment the passive obstruction of which has produced gross dilatation and hypertrophy of the colon above.

A rapid spurt of development followed his period of washouts: his voice broke, and he increased in height by 1 in. and gained 2 st. in weight in three months. Hæmoglobin rose to 100% without the administration of iron, and there was a great increase in general health and vigour. Pubic and axillary hair appeared within a few months (Fig. 3). Bowel washouts were continued at gradually increasing intervals for two years, but during the last year he has had a normal bowel habit and appears to need no treatment. He presents the appearances of a normal healthy young man with some very slight upper abdominal distension as the only abnormal sign. Secondary sex characteristics are normal.

Comment.—This man's story demonstrates the natural history of Hirschsprung's disease in the rare cases in which survival of early obstructive episodes allows the ultimate development of a normal bowel habit. He also shows well the characteristic radiological appearances of the non-functioning terminal segment of bowel, the passive obstruction of which leads to hypertrophy and dilatation of the colon above. His case demonstrates, too, the well-known association with dwarfism and sexual infantilism; this may be due to impairment of protein nutrition by compression and distortion of the small gut by fæcal masses in the colon.

There seems to be no present indication for operation or other treatment, though he may, in future, again need bowel washouts which he may be able to learn to carry out himself.

Acknowledgments.—I wish to thank Dr. H. S. Le Marquand, my colleague at Reading, for allowing me to maintain an interest in this case and to show him. Thanks are also due to Dr. E. H. P. Cave for the X-rays, to Dr. Sheldon for allowing me access to old notes and photographs, and to Mr. Denis Browne for his advice and interest.

Section of Laryngology

President—C. P. WILSON, C.V.O., F.R.C.S.

[*February* 5, 1954]

DISCUSSION ON TREATMENT OF CARCINOMA OF THE NASOPHARYNX

Professor Victor Lambert (Department of Otolaryngology, University of Manchester):

The clinical material on which this survey is based has been taken from the records of the Manchester Christie Cancer Hospital and Holt Radium Institute between the years 1932 and 1950.

The nasopharynx belongs to the province of the rhinologist, but as its limits are defined by structures which are of interest to other specialties, such as the orbit and its contents, the brain and its cranial nerves, much of our clinical material is seen first of all by the ophthalmologist, the neurologist and the neuro-surgeon. When nasopharyngeal malignancies present—as I hope to point out later on they so often do—by invasion of the cervical glands, patients seek the advice and help of the general surgeon before coming to us.

The post-nasal space is an area which presents very real difficulty for accurate examination: whether it is the post-nasal mirror which is used or the nasal endoscope there always are blind spots which are difficult to inspect. This is true not merely in the initial examination, when the diagnosis of a tumour is first being established, but also when the post-nasal cavity is to be inspected and assessed in the follow-up period after the treatment has been carried out. It is in both these directions that the work of our President, Mr. Paul Wilson, is so important, in that his palate fenestration technique proves so valuable permitting the direct and easy inspection of the pharyngeal cavity.

Surgery for malignant tumours of this region I regard as difficult and disappointing—in fact, as a generalization, I would say that surgery only has a place when it is "surgery of approach" to enable the radiotherapist to treat the primary tumour by X-rays or radium. I believe that the treatment of post-nasal malignancy is almost entirely their field.

Malignant disease of the nasopharynx occurs in something like 6 per million of our population. This figure is the estimate for its incidence in the North-West region of England. By the same method we have estimated that 30 per million suffer from cancer of the tongue. It is, therefore, much less frequent than tongue malignancy and slightly more frequent than malignancy of the palate.

The complicated embryology of the region results in tumours, both simple and malignant, of varying histology being found here. Most of the malignant tumours encountered fall easily into the following groups: (1) Carcinoma—with its subdivision into special sub-groups depending upon the cell type which constitutes the tumour; (2) The lympho-epithelioma; (3) The lymphosarcoma, and (4) The nasopharyngeal fibroma, which is regarded by most people as being only locally malignant, but it is probable that it does merge imperceptibly into the true sarcoma. This interesting tumour, with its almost predominantly male incidence and occurring round about puberty, is very rare, and both Mosher and other workers regard it as one of the diseases which are disappearing from human pathology.

The relative incidence of carcinoma and sarcoma of the nasopharynx is shown in Table I, taken from Ewing's list of 100 cases. It will be seen that 67% of these tumours are carcinomata. Another striking thing is the high percentage of lympho-epitheliomata—11%.

Table II shows that if the 32 "others" are taken out, there are 65% carcinomata.

Tables II–VI show an analysis of 132 tumours which I have collected from our records, and it will be seen that the relative proportion of carcinoma and sarcoma are much the same as those shown by Ewing.

Godtfredsen collected a series of 438 cases from Scandinavian clinics and of these 243 were carcinomata and 195 sarcomata.

Table V shows that in 22 of the tumours the pathology was never determined and also demonstrates the small number of lympho-epithelioma. The actual naming of the pathology of any given biopsy is bound to vary with different pathologists.

Fig. 1 shows the age incidence of these groups of tumours: a striking feature is the incidence of carcinomata in persons of middle age, that is, from 50 to 60, while the sarcoma figure appears to rise progressively, reaching its peak about 65. The difficulty in histological diagnosis of these tumours appears to be at its peak in the younger age groups.

Turning to the presenting signs and symptoms of nasopharyngeal malignancy (Fig. 2), the analysis of my series seems to show quite definitely that the commonest sign which brings the patient for treatment is the presence of enlarged glands in the neck—the glandular enlargement being far commoner as a presenting sign in the carcinomata than in the sarcomata, both actual and relatively. About a quarter of the cases of sarcomata announce their presence by secondary glands (9 out of 35) and a little less than half the carcinomata (28 out of 65) do so in like manner. Nasal symptoms appear to

be the next commonest warning of the presence of nasopharyngeal malignancy, nasal obstruction being about twice as common as epistaxis (28 to 15). Neuralgic pain of various types in this series is the third, and a fairly bad third single warning symptom (26). Deafness is not as common as one would think and was the presenting symptom in only 12 of the cases.

The question of pain as an initial symptom of post-nasal tumour has very important clinical implications. I am informed by Sir Geoffrey Jefferson, that a reassessment of the so-called primary tumours of the gasserian ganglion and the trigeminal nerve is called for, and in his Bowman Lecture (1953, *Trans. ophthal. Soc., U.K.*, **73**, 117) he expresses a view that many of these so-called primary tumours originate in the vault of the nasopharynx and spread from here into the middle fossa. In the same paper he makes two other important points—the frequency with which these tumours give rise to

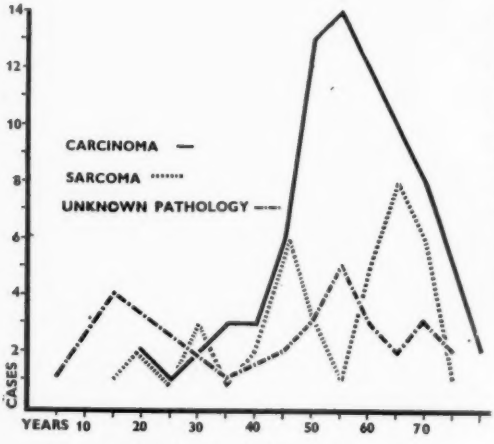


FIG. 1.—Age incidence.

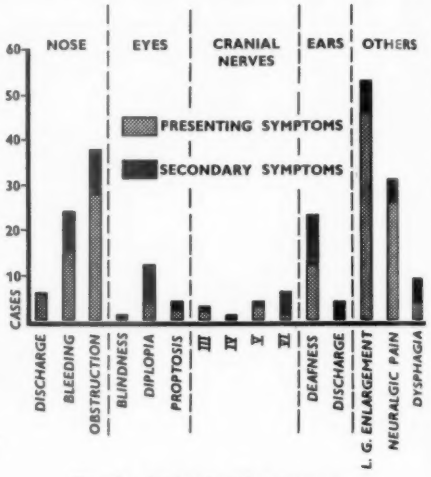


FIG. 2.—Incidence of symptoms.

TABLE I
Histology of 100 Cases (Ewing)

	Per cent
Squamous-cell carcinoma	30
Transitional-cell carcinoma	37
Lymphosarcoma	15
Lympho-epithelioma	11
Adenomamalignum	3
Adenoid cystic epithelioma	4

TABLE II
Gross Analysis of Post-nasal Tumours (Christie Hospital and Holt Radium Institute Series)

Carcinoma	65
Sarcoma	35
Others	32
	132

TABLE III
Christie Hospital and Holt Radium Institute 1932–1950
CARCINOMA (65)

Carcinoma (unclassified)	5
Undifferentiated	12
Squamous	20
Epidermal	7
Transitional cell	10
Anaplastic	10
Basal cell	1

TABLE IV
SARCOMA (35)

Lymphosarcoma	17
Reticulum cell	15
Round cell	1
Fibrosarcoma	1
Sarcoma	1

TABLE V
OTHER MALIGNANT TUMOURS (32)

Malignant melanoma	2
Mixed tumour	2
Juvenile fibroma	1
Glioma	1
Hypernephroma	1
Lympho-epithelioma	1
Pathology not determined	22
Malignancy uncertain	2

TABLE VI
Nasopharyngeal Tumours (132 Cases)

	Deaths	Well*
Carcinoma	55	15
Sarcoma	29	6
Unknown pathology	18	7
Juvenile fibroma	1	1
	103	29

*Five years after treatment.

secondary deposits in the neck glands, and the high incidence of mortality within the first two years of operation. I do not think that this whole statement is meant as a reproach to those who have some interest in the post-nasal space and its pathology, but it is, I believe, a serious warning, and stresses still further how, despite every care, a small primary tumour may be missed in the vault of the post-nasal space, and how important it is to have a basal projection X-ray picture taken of the base of the skull in all cases of neuralgia of the sensory nerves of the face.

All the cases in this series have been treated by X-rays or radium. It is our custom in Manchester for the surgeon to define, so far as is humanly possible, the site and limits of the tumour and to co-operate where the surgery of access is needed, but the details of technique and dosage is, in my opinion, the rightful province of the radiotherapist.

In trying to establish the accurate diagnosis before treatment is undertaken, a biopsy is the *sine qua non* of the diagnostic procedure, but those of us who have any experience of this region know only too well how hard it is for a pathologist to make an accurate diagnosis in many cases, and as will be appreciated from my figures, the percentage of unclassifiable tumours is fairly high.

Fig. 3 shows the heavy mortality of these cases in the first year or eighteen months, partly because, no doubt, many of the cases are so far advanced before treatment is sought.

Fig. 4 shows the survival of the 132 cases at the end of the five years. The carcinomata have probably the slightly better ultimate prognosis, though the initial mortality is pretty high.

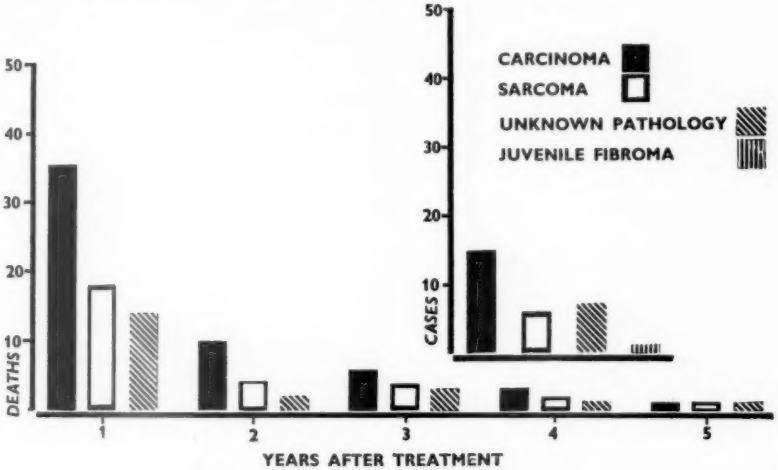


FIG. 3.—Mortality rate within 5 years after operation.
FIG. 4 (inset).—Patients well after five years.

Table VI shows the survival rate, the overall figure being somewhere in the region of 30% five-year survival. Whilst this might be regarded as a satisfactory figure in view of the nature and site of the disease, it gives no clue to the serious morbidity which accompanies the treatment of these patients, and it still remains one of the major problems in malignancy as it is related to our specialty.

Miss Margaret Snelling (The Meyerstein Institute of Radiotherapy, Middlesex Hospital, London :

The treatment of carcinoma of the nasopharynx is disappointing to the radiotherapist; the results are poor in spite of the fact that it is a disease which may be slow in evolution and which may remain localized to the head and neck, or even to the primary area for months or years.

This study is based on the 78 patients seen at the Middlesex Hospital between 1934 and 1953. Of these patients 76 were treated.

Table I shows the results of the treatment of 46 cases treated between 1934 and 1948. Of the 46 cases treated only 7 survived five years—a survival rate of 15·2%.

There were two main methods of treatment. Where there was no glandular involvement and where the growth was of relatively low malignancy small fields were used, the radiation was localized to the nasopharynx and a dose of up to 5,000 or 6,000 r was delivered over a period of four to six weeks. Where the glands of neck were involved or where the neoplasm was of such high malignancy that early spread was to be expected, the whole of the tissues from (and including) the base of skull to the clavicles were irradiated in one block. Because of the increased reaction due to the irradiation of such a large volume of tissue, the total dose was necessarily lower and varied between 2,500 and

3,500 r delivered in up to a month. Where a recurrence occurred at the primary site or in the glands it was re-treated with small localized fields. Such repeated treatments are usually less successful than the first, since the neoplasm is often less sensitive to radiation while the injury to the normal tissues is increased. We did not pursue treatment after the second full course. During the last years, however, we have not only been giving rather more intensive treatment in the first course but we have also been repeating the localized treatments several times in those slowly progressing cases where the disease is localized to the primary area or to that area and a limited number of glands. In many of these cases the recurrence of the primary growth was apparently small but signs of cranial nerve involvement showed there was deep infiltration. In these cases we have used a technique by which a high dose is delivered to a small volume of tissue. This necessitates very accurate localization of the growth and additional methods of diagnosis and of beam direction have been employed. Rather to our surprise we have found that the condition continues to respond to treatment, symptoms are relieved and there is often recovery of function of the cranial nerves. Some patients have received three or even four treatments to the nasopharynx each time with symptomatic relief but almost certainly without eradication of the disease. It is too early for full statistical evidence on the effect of the recent more intensive treatment but a comparison of the three-year survival rates for the years 1934–1948 and the years 1949 and 1950 shows a marked improvement in the latter group.

TABLE I.—FIVE-YEAR SURVIVAL, 1934–1948
New Cases, 1934–1948

	Total treated	Five-year survival
Histology proved ..	38	6 (15·8%)
All cases treated ..	46	7 (15·2%)
All cases seen ..	48	7 (14·6%)

TABLE II.—THREE-YEAR SURVIVAL RATE

	Cases treated 1934–1948		Cases treated 1949, 1950	
	Total treated	Three-year survival	Total treated	Three-year survival
Histology proved	38	9 (23·7%)	13	7 (53·8%)
All cases treated	46	10 (21·7%)	14	8 (57%)

Table II shows that the survival rate of the patients treated rose from 21·7% to 57%. While the number of cases in the recent series is low—14 against 46—the groups are in other ways comparable. Actually the recent series contains a smaller proportion of anaplastic carcinoma—the histological variety with the best prognosis—and 7 of the 8 survivors in this group showed evidence of glandular involvement before the beginning of treatment. It should, however, be emphasized again that these are survival and not cure figures and 3 of the 8 survivors are thought or known to have growth still present. The fact that this increase in survival can be obtained by intensive and repeated treatment has, however, induced us to review our technique in the hope of ultimately improving not only our survival rate but also the cure rate.

General Considerations

The effect of radiation on living tissues is always injurious, and since the volume irradiated necessarily includes normal tissue as well as tumour this also is injured. The effect on a cell is greatest when it is in process of division; a tumour cell is therefore more sensitive than is a normal cell and a highly malignant rapidly growing tumour is more sensitive than is a differentiated one. In radiotherapy advantage is taken of these different sensitivities and it is our aim to administer enough radiation to destroy the tumour cells without injuring the normal beyond possibility of recovery. The conservation of the normal tissues is especially important because the reaction of a tumour to radiation depends to some extent on the behaviour of the tumour bed. Success in radiotherapy depends upon a correct interrelationship between the volume of tissue irradiated, the quantity of radiation and the period of time over which it is given, and the response of each case is observed closely in order that the quantity of radiation should not be so high as to cause too severe a reaction nor so low as to fail to destroy the tumour. Various methods are used to increase the tolerance of the normal tissues in relation to that of the tumour. The overall duration of the treatment may be lengthened or the volume to be irradiated diminished while the use of heavily filtered deep X-rays, of supervoltage X-rays or of the gamma rays of radium or of radioactive cobalt also reduces the reaction produced in the normal tissues.

Extent of the Disease

The diagnosis of these tumours is often late owing to the fact that frequently there may be no symptoms referable to the primary lesion and that the region is relatively inaccessible to thorough examination.

Table III shows the incidence of glandular involvement in 46 cases treated between 1934 and 1948. 4 of the 14 patients without glandular involvement survived for five years. Where the glands are involved and the volume to be irradiated extends from the base of skull to the clavicles success can only be achieved in radiosensitive lesions. While 3 of 16 cases of anaplastic carcinoma and lympho-epithelioma with glandular involvement survived five years none of the 6 keratinizing squamous-cell carcinoma and transitional-cell carcinoma survived for the same period.

Some improvement might follow an increase in the total dose given to the neoplastic tissue. This could be achieved by an increase in the total duration of the treatment or by the use of heavily filtered

deep X-rays, supervoltage X-rays or gamma rays. The use for part of the time at least of multiple small fields also increases the possible dose but may be impossible because of the anatomical distribution of the glandular metastases.

TABLE III.—GLANDULAR INVOLVEMENT
46 New Cases Treated 1934–1948

	Total treated	Five-year survival
No glands	14	4
Glands involved ..	28	3
Details not available ..	4	

TABLE IV.—HISTOLOGY
Carcinoma of the Nasopharynx 1934–1948

	Total treated	Five-year survival
Anaplastic carcinoma ..	20	4
Differentiated carcinoma ..	10	1
Lympho-epithelioma ..	7	1
Transitional-cell carcinoma	1	0
Total	38	6 (15·8%)

Response of the Primary Growth

In all 76 cases have been treated in the Department from 1934 up to the end of 1953. Of 44 of these patients who died under observation, active growth was present at the primary site in 24, 12 of whom showed evidence of extension to the base of the skull with involvement of the cranial nerves. Failure to destroy the primary growth may be caused by a variety of factors—it may be histologically of a radioresistant type or there may be local factors such as poor blood supply or involvement of bone or cartilage which decrease its natural sensitivity to radiation. The dose or dosage rate may be wrongly chosen or the localization of the treatment may be faulty.

Table IV shows the result of the treatment of various histological types. While rapidly growing tumours such as the lympho-epitheliomata and the most malignant types of anaplastic carcinoma respond quickly to small doses of radiation, glandular involvement is early and they may spread beyond the treatable area before the requisite dose has been given. At the other end of the scale are the slowly growing differentiated tumours, which metastasize late but are, unfortunately, relatively radioresistant. The best results are obtained in the medium group of cases such as the less malignant anaplastic carcinomata which are active enough to respond well to treatment but are not so malignant as to metastasize early.

TABLE V.—CARCINOMA OF NASOPHARYNX
Cases Without Recurrence of Primary Growth

	Dose	Duration of treatment	Progress
Anaplastic carcinoma			
No glands	5,000 r	59 days	AW 11 years
	5,920 r	40 ,,	AW 9 ,,
	5,481 r	46 ,,	AW 3 ,,
Unilateral glands	4,300 r	42 ,,	AW 9 ,,
	6,625 r	62 ,,	AW 4 ,,
Bilateral glands	2,050 r	18 ,,	AW 9 ,,
Differentiated carcinoma			
No glands	7,614 r	44 ,,	DOC 5½ ,, (no rec.)
Lympho-epithelioma			
Unilateral glands	6,629 r	66 ,,	AW 3½ ,,
	3,475 r	40 ,,	AW 3½ ,,
No biopsy			
No glands (cranial nerves inv.) ..	4,501 r	38 ,,	AW 6 ,,
Bilateral glands	4,428 r	48 ,,	AW 4 ,,

Table V shows the dosage and time used in the treatment of a series of cases who have remained free from recurrence over periods of three or more years. It will be seen that although the majority of cases are of radiosensitive lesions, in all but 2 cases the doses used are higher than those that can be obtained by the large field technique and vary between 4,300 and 6,600 r delivered over a period varying between 38 and 66 days. Only one case has been cured with a dose as low as 2,050 r which was delivered by large opposing fields over 18 days. From these figures it appears that improvement in the results could be achieved by increasing the total dose and to do this the treatment time must be extended and the amount of normal tissue included in the irradiated area should be reduced to a minimum. Very accurate localization of the tumour is essential and this has necessitated a series of clinical and anatomical investigations to be described by Mr. Flatman. Post-mortem investigations will doubtless help but none have been possible since the 5 or 6 cases with cranial nerve involvement who have been seen since this work was started have all responded to repeated treatment.

Clinical examination includes full examination of the cranial nerves, X-rays are taken to show the nasopharynx and base of skull and where no lesion is obvious on post-nasal examination but a hidden primary is suspected, a transpalatal exploration is carried out and a biopsy is taken from

the mucosa on the roof and the lateral recess of the nasopharynx. If the diagnosis is confirmed, the present practice is to convert the palatal incision into a permanent palatal fenestra before treatment is commenced (Wilson, 1951). The major portion of the hard palate is removed together with the posterior part of the nasal septum and the posterior ends of the inferior turbinates—the removal of the latter facilitates inspection of the mucosa in front of the eustachian orifice and makes possible the use of an intracavitary applicator should this prove necessary. That technique of treatment is then chosen which is considered to be best adapted to the clinical condition.

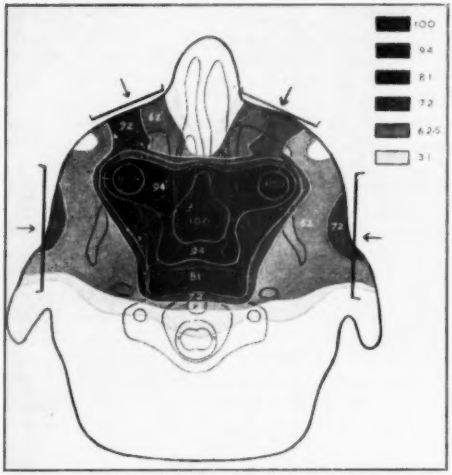


FIG. 1.—Dose distribution obtained in treatment of nasopharynx by 4-field technique using 250 kV X-rays and treatment distance of 40 cm.

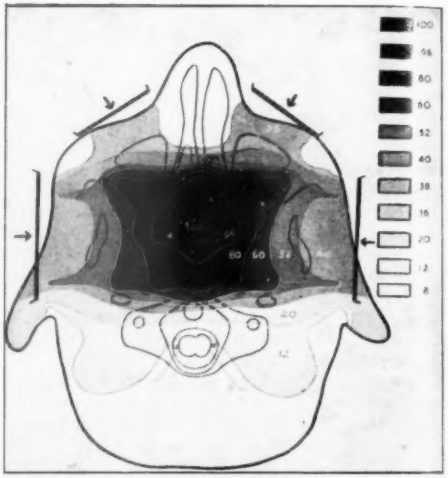


FIG. 2.—Dose distribution obtained in treatment of nasopharynx by x-field technique with source of radioactive cobalt. Treatment distance 80 cm.

Fig. 1 shows the distribution obtained by a conventional 4 field treatment using deep X-rays generated at 250 kilovolts and treating at a distance of 40 cm. Using this technique a dosage of 5,500–6,000 r is administered over a period of five to six weeks to the central area; the skin and normal tissues are also irradiated fairly heavily, although more than slight irradiation to the orbits and spinal cord is avoided. Such treatment causes moderate scarring of the normal tissues but should recurrence occur and treatment be repeated, the scarring becomes very considerable.

Fig. 2 based on the depth dose data published by Harold Johns of Saskatoon (Fedoruk, Johns and Watson, 1953) shows the same arrangement of fields, using a large source of radioactive cobalt, equivalent to about 1,500 grammes of radium, the source skin distance being 80 cm. By the use of this type of radiation, and especially of the increased treatment distance made possible by the enormous power of the source, the area of high dosage becomes more localized and the normal tissues receive much less irradiation.

A comparison of these two charts demonstrates the advantage to be gained by the use of powerful sources of penetrating radiation such as the cobalt unit or supervoltage X-rays. A 10-gramme tele-radium unit cannot have the same effect since the source is not strong enough for the treatment distance to be increased beyond 8 or 10 cm. If this distance were exceeded the depth dose would be improved but the time taken would increase to such an extent as to be quite impracticable.

The distribution in these techniques can be varied to some extent when dealing with different cases and Fig. 3 shows how the addition of two postero-lateral fields pulls back the area of high dosage so as to give more radiation to the structures at the posterior part of the nasopharynx.

In addition to supervoltage irradiation the problems of dose distributions have been overcome to some extent by the use of rotational techniques. One of these methods is by the use of the Siemens Convergent Beam unit. Its effect is produced by continuous movement of the X-ray tube through a spiral path. The tube is set at such an angle that the beam is directed throughout the treatment at a preselected spot at a known depth. By this means each part of the skin and superficial tissues is irradiated during only a fraction of the time but the tumour is treated continuously.

Fig. 4 shows a typical dose distribution from the convergent beam, and it can be seen that the area of high dosage is very small. This method is therefore only suited for patients with very localized disease, and we have used it chiefly for re-treatments when a recurrence has developed after a 4 or 6 field treatment. In such cases it has proved extremely useful since the dose received by the normal tissues is very low.

Fig. 5 shows the dosage distribution of the same treatment drawn on a sagittal section of the head and it is obvious that not only must the radiotherapist's knowledge as to the site of the tumour be accurate, but also great accuracy must be used in the direction of the beam. This can be checked by radiographs taken with the therapy tube when the patient is in the treatment position.

Another high dose small volume technique involves the use of an intracavitary applicator, inserted through the fenestra and loaded with a source of radioactive cobalt or with a radium needle.

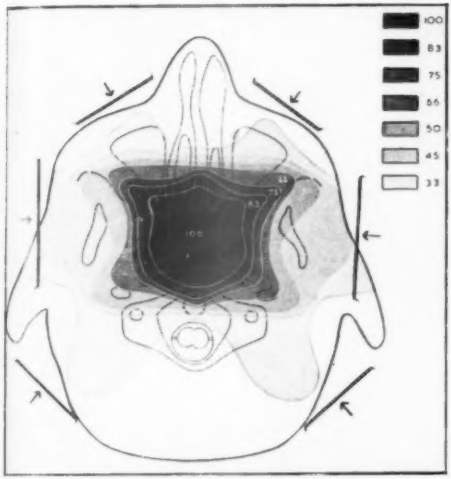


FIG. 3.—Dose distribution obtained in treatment of nasopharynx by 6-field technique with source of radioactive cobalt. Treatment distance 80 cm.

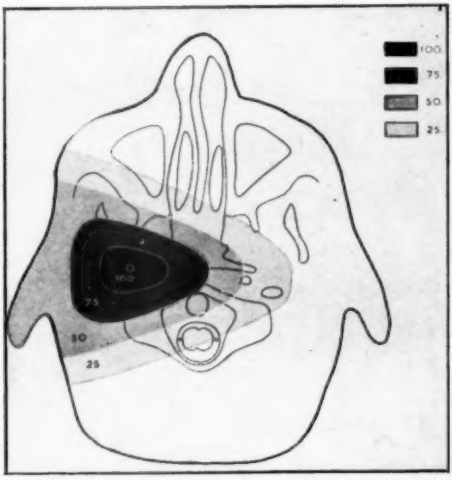


FIG. 4.—Dose distribution obtained in treatment of nasopharynx using Siemens Convergent Beam unit.

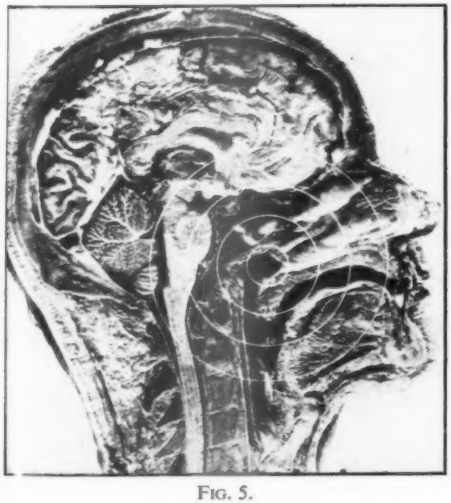


FIG. 5.

FIG. 6.

FIG. 5.—Dose distribution obtained in treatment of nasopharynx using Siemens Convergent Beam unit.

FIG. 6.—Intracavitary applicator containing small source of radioactive cobalt.

In such an applicator (Fig. 6), the source is mounted on a block of lead which prevents too high irradiation of the palate. The advantage of this method is the localization of the high dosage to a small volume of tissue—the corresponding disadvantage the rapid fall off of the dose, which diminishes with the square of the distance from the source.

Fig. 7 shows a sagittal section of a head in which a fenestration has been performed and an applicator has been placed in position. The circles show clearly how rapidly the dose diminishes—thus if at the surface of a growth 1 cm. from the source the dose is 6,000 r, the dose 1 cm. below the surface is only 1,500 r—far too low for the treatment of carcinoma.

Fig. 8 shows a transverse section of the head with the source in position and from it can be seen again how the treatment is limited always by the dosage received by the tissues nearest to the source; for this reason it is useful only for those cases where the neoplasm is on a projecting part of the nasopharynx—on the roof or posterior wall—and it is unsuitable for any case where the growth is infiltrating or where it is in the lateral recess. If localized treatment is necessary, such cases should be treated with the convergent beam unit or by some other form of rotation treatment.

In conclusion I should like to quote two case histories which show some of the difficulties which arise in the treatment of these cases.

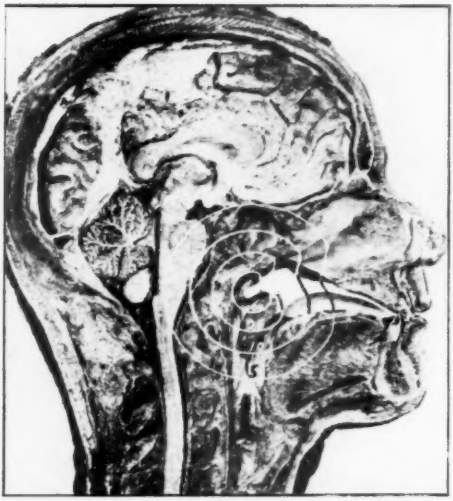


FIG. 7.—Dose distribution obtained in treatment of nasopharynx using intracavitary applicator.

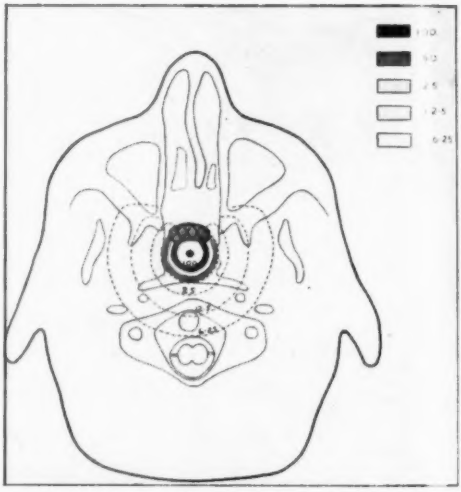


FIG. 8.—Dose distribution obtained in treatment of nasopharynx with intracavitary applicator.

Case I.—Lympho-epithelioma of left side nasopharynx with bilateral glands.

August 1951: Deep X-rays 3,500 r in 40 days (large fields). Primary and glands disappeared.

June 1952: Recurrence of primary and glands. Deep X-rays 5,700 r in 34 days (small fields).

August 1952: Recurrence on right side of nasopharynx. Fenestration and convergent beam therapy (6,400 r in 35 days to post-eustachian recess).

March 1953: Involvement of left V and VI nerves. Exploration of sphenoidal sinus by Mr. C. P. Wilson showed invasion of left wall. Convergent beam therapy 6,000 r in 38 days.

Total roentgens delivered over seventeen months—22,000 r.

Maximum dose to any part of nasopharynx—16,000 r.

Case II.—Enlarged glands right side of neck. No obvious primary.

June 1951: Deep X-rays to glands (local fields).

April 1953: Left-sided pain and deafness. Small carcinoma on roof of nasopharynx. Fenestration and treatment by intracavitary applicator (8,000 r to base of ulcer in 27 days).

June 1953: Left V nerve pain and VI nerve palsy, followed by involvement of IX, X, XI and XII nerves. Convergent beam therapy 6,000 r in 34 days.

November 1953: Primary growth healed, recovery of function of cranial nerves. Deep X-rays to glands right side of neck.

Each one of the cases we have treated during the last few years has presented similar but individual problems and to the extent that there has been an increased rate of survival, we feel that our technique has improved. On the other hand it is impossible to be complacent with the results of treatment in which the improvement appears to be of only temporary duration. It is obvious that we are only on the fringe of the problem, and that further research is necessary if we are to improve our methods of treatment.

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Mr. G. E. Flatman (The Meyerstein Institute of Radiotherapy, Middlesex Hospital, London):

The primary site of tumours of the nasopharynx and their tendency to infiltrate the base of skull and penetrate the cranial cavity are of particular interest in the development of techniques of irradiation designed to deliver high dosage to relatively small volumes of tissue. A photographic method of beam direction using the X-ray therapy tube under treatment conditions is in common use as a means of localizing these volumes. It has, however, been realized that our detailed knowledge of the anatomical relations at the base of the skull and vault of nasopharynx is subject to considerable limitations and inaccuracy. However, the common occurrence of cranial nerve involvement by tumours arising in the nasopharynx has led to further considerations of the cranial nerve roots and main trunks in particular relation to this region, and it is suggested that a clearer understanding of the anatomy will contribute towards a more accurate localization of the invading tumour.

Reference to the extensive literature reveals, amongst others, the names of Gradenigo, Trotter and Ballenger associated with intracranial nerve syndromes and Tapia, Vernet and Burger with extracranial nerve syndromes. It is not always clear how the nerves become involved by the particular disease, though in the case of nasopharyngeal tumours arising in the region of the fossa of Rosenmüller, Jacod pointed out their tendency to track alongside the eustachian tube and penetrate the foramen lacerum. Godtfredsen in his supplement to *Acta Otolaryngologica* (1944) points out that in only a scant one-third of cases in which one or more cranial nerves are involved either intra- or extracranially is there radiographically demonstrable erosion of the base of the skull. Clearly, therefore, in a good proportion of cases the tumour penetrates the skull in the first instance through one or more of the preformed foraminæ including the foramen lacerum.

A review of the 78 cases on which Miss Snelling's report is based shows that 30 had cranial nerve involvement. Of these 12 had radiographic evidence of erosion of the base of the skull at some stage of the disease.

The variety and number of combinations of nerves affected, as shown in Fig. 1, was so wide that it would be inaccurate to regard any one of them as a syndrome.

The order in which the combinations appeared was variable, but with few exceptions the initial presentation was of pain referable to the maxillary division of the V cranial nerve. The VI cranial nerve was next in order of involvement which was nearly as common as that of the V (Fig. 2).

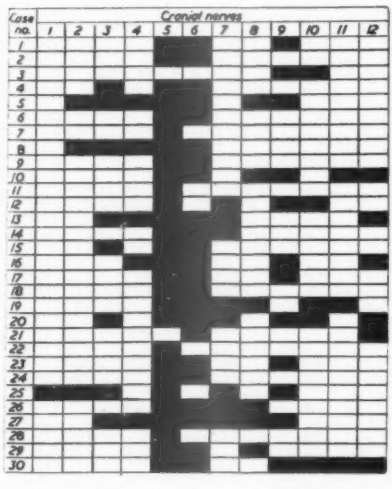

FIG. 1.—This shows the combinations of cranial nerves involved in each of 30 cases of carcinoma of the nasopharynx.

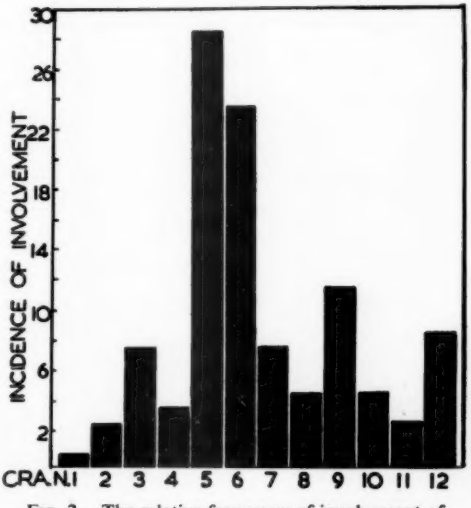

FIG. 2.—The relative frequency of involvement of each cranial nerve in the 30 cases.

I shall later show that if the relevant clinical manifestations had been looked for, involvement of the nerve of the pterygoid canal might well have been reported in a number of cases.

Despite my conclusions that combinations of cranial nerve palsies do not represent reproducible syndromes, their presence and the order of involvement may prove a valuable aid to the localization of tumour in each individual case. It is not intended to postulate any specific mode or direction of tumour spread nor is it considered that we are necessarily commonly dealing with small volumes of intracranial tumour.

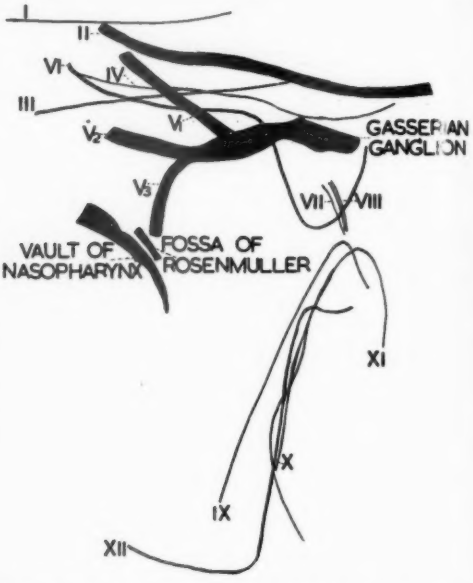

FIG. 3A.—Lateral radiograph of section of skull in which the cranial nerves (main trunks) have been replaced with wire.

FIG. 3B.

F.G. 3C.

The association of radiographically demonstrable erosion of the base of the skull is of particular value in the localization of tumour where the number of cranial nerves involved is few or nil. It is surprising how large an area of erosion may be found on first examination and there appears to be little relationship between the area of erosion and the number of nerves involved.

Further studies of the anatomy of the nasopharynx and base of skull have been made. Radiographic examination of a section of head in which the cranial nerves had been dissected out and replaced by wire of identical gauge demonstrates their relationship to the vault of the nasopharynx and the fossa of Rosenmüller (Fig. 3 A, B, C).

In the region of the cavernous sinus where it is presumed the V and VI nerves are involved, the greater superficial petrosal nerve is seen lying deep to the mandibular division of the V nerve before it is joined by the deep petrosal nerve to form the nerve of the pterygoid canal.

FIG. 3B.—A.-P. radiograph of section.

FIG. 3C.—Submento-vertical radiograph of section.

This nerve is therefore closer to the nasopharynx than any other above the fossa of Rosenmüller (Fig. 4).

Lateral dissection in the neck shows the relationship of the nerve of the pterygoid canal and the eustachian cartilage with the levator palati muscle partly taking origin from it after the lateral pterygoid plate has been removed and the tensor palati muscle reflected forward (Fig. 5).

Reference to a sphenoid bone and to a coronal section of head through the region of the eustachian tubes shows the nerve of the pterygoid canal, maxillary nerve and abducent nerve in a direct line above the fossa of Rosenmüller.

From these studies it is concluded that the most likely course of a tumour spreading up into the skull from the region of the fossa of Rosenmüller is either through the foramen lacerum or by erosion of bone in close proximity to it, including the floor of the sphenoid sinus. The most likely order in which the cranial nerves will be involved, therefore, will be first of all the nerve of the pterygoid

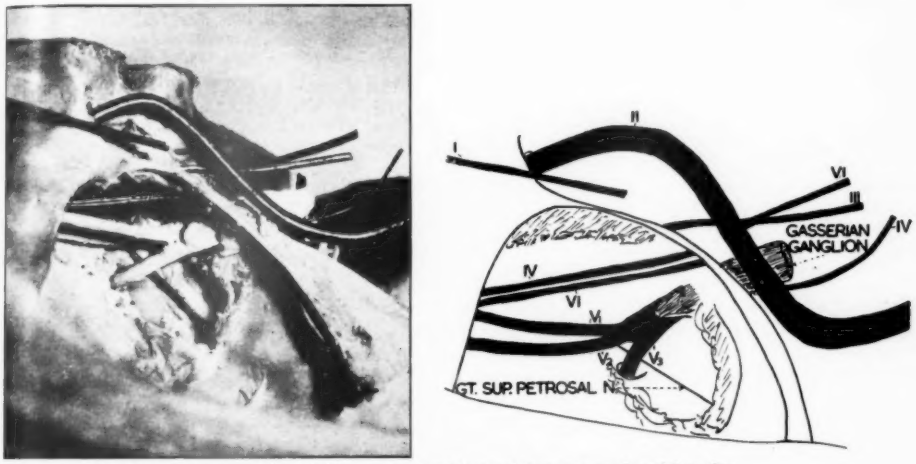

FIG. 4.—Cavernous sinus laid open and with cranial nerves replaced by wire.

canal and then the maxillary and abducent nerves. The occurrence in that order of involvement of the latter two nerves is borne out by clinical experience and by statistics. Unfortunately, however, it was not until recently that it was appreciated that the nerve of the pterygoid canal is by far the

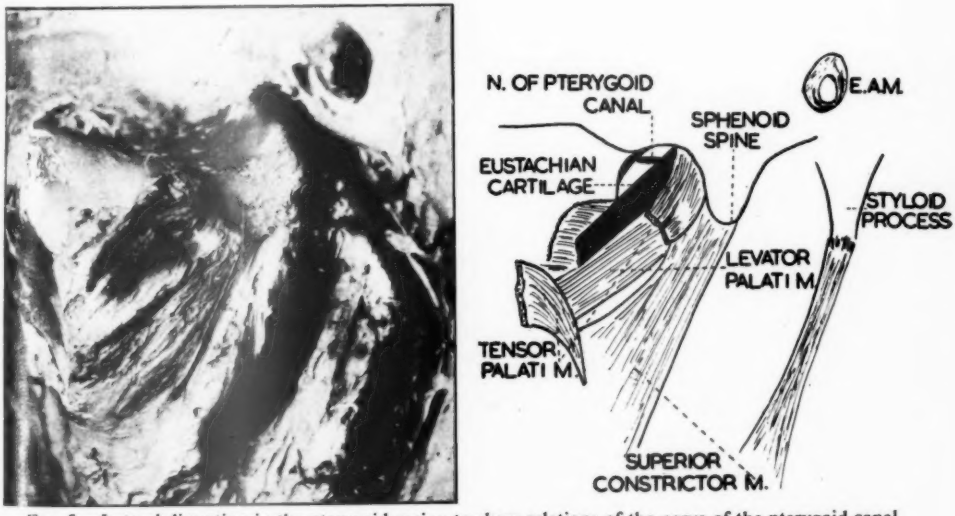

FIG. 5.—Lateral dissection in the pterygoid region to show relations of the nerve of the pterygoid canal.

closest to the fossa of Rosenmüller. Past clinical records are not sufficiently accurate in respect of detailed descriptions for us to name symptoms referable to this nerve such as a disturbance of lacrimation as preceding maxillary nerve symptoms. This is regrettable.

It is quite obvious that when the stage of VI nerve palsy is reached the tumour has already spread at least 2 cm. from its point of origin, and even further when other cranial nerves are involved.

This detailed investigation suggests a means of making an earlier diagnosis of the spread of tumours of the nasopharynx, particularly in the difficult group of cases with neurological signs and little or no evidence of any abnormality in the post-nasal space. An appreciation of the anatomical relationships has suggested an extension of the transpalatal exploration into the formation of a permanent fenestra. The technique of this operation has previously been described by Mr. C. P. Wilson. This has been of some value for the purpose of use of a local source of irradiation to high dosage, although in practice it has proved disappointing because of the impossibility of obtaining an adequate depth dose from a source so close to a tumour. It has, however, proved of value in enabling observation of local reaction to treatment, and does provide, without disability to the patient, a means of inspection of that elusive area, the nasopharynx. Unfortunately this again is disappointing because several recent patients have shown evidence of active growth by reason of cranial nerve involvement without having any visible growth in the nasopharynx. The possibility of providing something better by surgical means is now being considered, and Mr. Wilson has recently explored the roof of the nasopharynx with a view to assessing the extent and position of recurrence of tumour in such a case. From this it is felt that possibilities are held out which will help towards a solution of the problem of improved treatment of base of skull infiltration.

ACKNOWLEDGMENTS

I wish to thank Professor Windeyer, in whose department I work, and Mr. C. P. Wilson for their encouragement and assistance in carrying out this investigation. In addition I would like to acknowledge the facilities placed at my disposal by Sir Harold Graham Hodgson and Professor E. W. Walls.

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Professor E. W. Walls, at the invitation of the President, gave a blackboard demonstration of the relations of the pharyngeal recess. Although formerly believed to be a remnant of the second pharyngeal pouch this view was no longer held, and the recess was now considered to be formed as a secondary extension of the primitive pharynx. The depth of the recess was variable and possibly in some cases its lateral portion became obliterated during development. Laterally it extended below the foramen lacerum and petrous temporal where it lay close to the nerve of the pterygoid canal (vidian nerve). Pressure on this nerve by a tumour could be expected to affect its secreto-motor and taste fibres with the production of symptoms referable to the lacrimal gland and palate. To an anatomist the whole subject was very refreshing, illustrating as it did the importance of detailed topographical anatomy in the early diagnosis of a very grave condition.

Dr. M. Lederman reported his experiences at the Royal Cancer Hospital.

Table I shows some of the salient features of the clinical material seen. Table II shows the histological types of tumour encountered and the results obtained.

Dr. Lederman then went on to say that the relatively favourable results obtained in the sarcoma group should not blind one to the poor results obtained in the squamous carcinoma group: progress in radiotherapy should be assessed in relation to improvement in these latter results.

The following features of clinical interest were noted:

(1) Loss of the corneal reflex was sometimes the earliest sign heralding invasion of the base of the skull.

(2) When following-up patients treated for nasopharyngeal neoplasms attention should not be limited to the head and neck. A general examination was also required since one in four of all patients in this series develop distant metastases.

(3) In the presence of cranial nerve involvement spread through the cerebral spinal axis was possible without obvious invasion of the base of the skull or basal meninges. Entry into the sub-arachnoid space took place presumably via the nerve sheaths or by the perineural lymphatics, and in these circumstances it was not unusual for patients to develop root pains without radiologic evidence of secondary deposits; in one such case a paraplegia developed.

TABLE I.—CLINICAL MATERIAL

The Royal Cancer Hospital, London
The Royal National Throat, Nose and Ear Hospital, London
CA. NASOPHARYNX—1933–1953

Number of cases seen	132	Sex: M 88 F 33
Number of previously untreated cases	121	Age: Youngest 4 years
		Oldest 84 years
Number of recurrent cases	5	70% Between 50 and 80 years
Number of cases seen but not treated by radiotherapy	6	14% Below 30 years
Number with lymph-node metastases when first seen	91/121 (75%)	
Number with cranial nerve lesions when first seen	34/121 (28%)	

TABLE II.—RESULTS IN RELATION TO HISTOLOGICAL TYPES
CARCINOMA NASOPHARYNX

Histology	Number of cases	Survival 5 year	4 year	3 year
Squamous epithelioma	74/121 (61%)	5/32 (16%)	8/35 (23%)	9/43 (21%)
Reticulosarcoma	6	2/4	2/4	2/4
Lymphosarcoma	8	1/6	2/7	2/7
Lympho-epithelioma	6	2/4	5/5	5/5
Transitional cell ca.	2	–/1	–/1	–/1
	22/121 (18%)	5/15 (33%)	9/17 (53%)	9/17 (53%)
Fibrosarcoma	2			
Rhabdomyosarcoma	1			
Lyposarcoma	1	5/121	1/2	1/2	1/2
Myxosarcoma	1			
Adenocarcinoma	2			
Salivary gland tumour	1	4/121	None available for three-year follow-up	
Chordoma	1			
Anaplastic malignant tumours	7/121 (6%)	None available for three-year follow-up		
No positive section	9/121 (7%)	–/7	1/8	1/8
Total	121	11/56 (20%)	19/62 (31%)	20/70 (29%)

Mr. Myles Formby said that when discussing the absence or presence of glands one must bear in mind the difference between the macroscopic and microscopic spread of the tumour. It was easy to remove macroscopically operable glands, but it had to be remembered, as Professor Windeyer had remarked earlier, that there were not only many areas inaccessible to surgery but also cells scattered through lymphatic channels. There could be no doubt that irradiation before operation might be extremely helpful in improving the surgical results.

Mr. Robert Owen emphasized the importance of assessing correctly the clinical signs with the anatomical involvement. He would like to know how often the transpalatal approach was carried out in order to assess the degree of spread and would this be done for the purpose of a biopsy only or would a fenestra be produced in order to facilitate radiotherapeutic treatment?

The other problem was the treatment of early palpable nodes. If the biopsy of the nasopharyngeal mass was returned as a squamous carcinoma it was his practice to carry out a block dissection first and then refer the primary for radiation. It was unusual for early operable nodes to be bilateral. If the other side became involved later another block could be performed. He did not believe in relying on radiotherapy in early operable nodes when the primary proved to be a squamous carcinoma unless it was very anaplastic. On more than one occasion he had found the recurrence of nodes in the neck after radiation a hopeless surgical problem but not when first seen before radiation.

Professor B. W. Windeyer said that all malignant tumours of the nasopharynx should not be placed in a single category and the method of treatment considered on the basis of whether metastatic nodes were technically operable or not. Some of the growths were carcinomas of only moderate

radiosensitivity, but others, including the whole group which had been classed by Dr. Lederman as sarcomas, were more radiosensitive and their metastases could be made to disappear by radiotherapy alone. Secondly, although he thought there might often be value in the surgical removal of a metastatic mass of radioresistant tumour, he doubted whether the metastases in lymph nodes from tumours in the nasopharynx were ever operable in the sense that the disease could be completely removed by block dissection, because the highest node involved was in the retropharyngeal region and not in the deep cervical group. He quoted a recent patient who at autopsy was found to have median retropharyngeal glands involved by cancer both at the level of the eustachian cushion and at the cricoid cartilage.

Mr. Robert Owen said that the case mentioned by Professor Windeyer was a rare picture. It was like speaking of a secondary node in the tonsil with a primary in the kidney . . . which could happen—he had seen such a case.

There was nothing like a block dissection to improve the cure rate.

The President said with regard to glands he was against radical surgery as a principle although in practice there were occasions when it was desirable. These patients did not as a rule die from their secondary glands but from extension of the primary growth. The general treatment was radiotherapeutic and not surgical and the glands could be treated as part of the planning of treatment for the primary tumour. A preliminary block dissection introduced disadvantageous complications into the radiotherapy planning and was, in any case, unnecessary.

Many cases came along having had a local excision of a gland for purposes of diagnosis before the nasopharynx had even been examined and this seemed quite wrong in principle.

Glands might be removed at a later date if they persisted after treatment had apparently got rid of the primary tumour but surgery in treatment of carcinoma of the nasopharynx was an ancillary to radiotherapy and not an alternative.

The trend of treatment at present was to cover a wide area primarily and then to make one's main attack on the primary growth with as localized a field as possible. This was the reason for stressing early diagnosis on the one hand and as accurate an assessment of the area involved on the other.

Mr. Flatman had rightly pointed out that the vidian nerve should be involved before the maxillary division of the V or the abducent and one should be very suspicious of carcinoma with merely increased lacrimation and a diminished corneal reflex on one side in a person past middle age. If to these were added persistent maxillary nerve pain then there was justification for doing a biopsy of the posterior wall of the lateral recess of the pharynx via a transverse palatal exploration.

Fenestration of the hard palate involved the minimum of discomfort or disability and was extremely useful in enabling observation to be carried out during subsequent treatment. It was now done as a routine at the Middlesex as the first stage in treatment.

Lately, investigation of the base of the skull had been carried out through the fenestration after treatment had been completed, if there were a recurrence of nerve palsies. It was amazing how great an area of involvement of the base of the skull was found when the nasopharynx appeared quite healthy.

The area of disease was always much greater than local appearances suggested and greater also than appeared on radiological examination.

In a recent case bone was found to be involved almost as far back as the margin of the foramen magnum.

It was hoped that more would be learned by the efforts being made to localize as accurately as possible the extent of the growth. There might be a place for intracavitary radiation following extensive exploration of the base of the skull in the same way as was done for carcinoma of the upper jaw.

Lastly it must be stressed that a large proportion of these cases were of comparatively low malignancy and even five-year survival statistics did not mean that the primary tumour had been eradicated.

Section of Epidemiology and Preventive Medicine

President—A. BRADFORD HILL, C.B.E., D.Sc.

[*November* 20, 1953]

Immunological and Epidemiological Problems

By THOMAS FRANCIS, Jr., M.D.

Henry Sewall Professor of Epidemiology, School of Public Health and Medical School University of Michigan, Ann Arbor, Michigan

FROM the time that antigenic variation among strains of influenza virus of human origin was first established, two general features have stood out. The first is that distinct variants have presented themselves from one year to another; this has led to a thesis that there is a progressive variation of strains "in one direction" with older antigens falling out and being replaced by strains with completely new antigenic components. The second is that while differences among strains can be detected there are extensive cross-relationships indicating the importance of common antigenic composition.

Studies in the Virus Laboratory at the University of Michigan have resulted in two new bodies of data which are of significance in understanding of the entire problem of strain variation and its relation to immunity and preventive vaccination. The reports have since been published.

In the one investigation (Jensen and Francis, 1953) the antigenic composition of influenza virus was measured by antibody absorption, employing inactivated virus attached to human erythrocytes in a stable preparation for absorption. It has provided a more comprehensive demonstration of strain differences and relationships. There have been clearly disclosed at least 18 measurable antigenic components in Type A strains and the procedure has permitted the simultaneous detection of variable quantities of different antigens in a strain so that a profile of a strain's constitution is provided. The wide sharing of components among strains indicates that no completely new antigens have formed in a given year nor do the components of strains from one year completely disappear in the next succeeding variants. Most strains possess a large number of antigenic components and the type of alteration observed appears to represent a rearrangement with certain of them dominating the character of one strain, while others which are present to lesser degrees in some strains, may be dominant in others. It was demonstrated that all antibody against the entire range of demonstrated antigens could be removed by combination of 5–7 strains selected because of their antigenic make-up. The outlook for increasingly effective vaccination is brightened by the possibility of making such a combination for vaccine to cover the entire range.

The second investigation (Davenport *et al.*, 1953) deals with the effects on the antibody content of the population which result from repeated exposure to antigenic variants of influenza viruses. It was studied by measuring, with many strains, the antibody content of lots of gamma globulin prepared in different years and the patterns of antibody found in sera collected in one year from various age groups.

In all samples of gamma globulin collected from 1943 through 1951, high levels of antibody were found with strains of Type A and Type B influenza viruses isolated prior to 1941. The highest levels were found in the more recent collections of gamma globulin. Antibodies to A-prime, and to B strains of 1945 and 1953, were present at low levels in gamma globulin collected prior to the isolation of these viruses. A moderate increase in antibody was observed in the gamma globulin of recent years.

The pattern of distribution of antibody by age in serum pools, exhibited high levels in infancy and childhood with most A-prime strains, but after the age of 20, little or no antibody was detected. With Type A strains antibody was usually not observed until the 11th year of age. Thereafter, high levels were present until age 20, when the amount of antibody declines to a moderate and relatively constant level which persists throughout life. Antibody against swine influenza virus did not become detectable until the 29th year. The intermediate antigenic character of a few A-prime isolates was reflected in the antibody pattern obtained with them. Antibody was not found until age 13 with the Lee (1940) strain of Type B influenza virus, but thereafter the level was high. With the Type B isolates of 1945 and 1952, antibody became measurable at earlier ages.

The present data clearly demonstrate that in the early years of life the range of the antibody spectrum is narrow, and that it becomes progressively broader in later life.

A striking correlation was found between what is known of the periods of prevalence of certain strains of influenza viruses and the age of the people in whom strain-specific antibodies are currently found. The age at which antibody to a given strain begins to appear in the population and the number of years during which strain-specific antibody levels are high indicate the time of prevalence of strains of that general antigenic character and also mark major shifts in the dominant antigens of influenza viruses. Thus by determining antibody patterns in human sera at any period of time, a serologic recapitulation of past prevalence of various strains of influenza virus is obtained. For example, the data clearly indicate that swine influenza virus possesses a major antigen of the virus involved in the pandemic of 1918. This interpretation follows the observation that the peak titre

in the serologic pattern to swine virus is now observed at age 35 to 38, and the levels are high at all ages above 32 years. The persons with highest titres are obviously those who were young children in the 1918 period, the group which experienced the greatest incidence of pandemic influenza. Moreover, the data make plain that strains of virus antigenically similar to swine virus and the virus of 1918 pandemic were probably prevalent before and after 1918, since the antibody level is high for over a decade, and some of the persons showing antibodies to swine virus were unborn in 1918. The possibility that swine influenza virus was that of the 1918 human epidemic was originally suggested by Laidlaw and by Shope. The present data which point out the shift in age incidence of antibodies to that strain support their hypothesis.

From these data the following immunologic thesis is formulated. The antibody which is acquired during the initial infections of childhood is of limited scope and reflects the dominant antigens of the prevailing strains. The immunity conferred by the initial experiences with influenza is also limited. Successive experiences later in life with viruses of related but differing antigenic make-up result in a composite of antibody which is oriented toward a larger number of the common antigens which comprise influenza virus. These experiences confer a broader immunity which limits infection with, and antibody response to, the more recently encountered strains. The antibody-forming mechanisms appear to be oriented by the initial infections of childhood so that exposures later in life to antigenically related strains result in a progressive reinforcement of the primary antibody. The highest cumulative antibody levels detectable in a particular age group tend, therefore, to reflect the dominant antigens of the virus responsible for the childhood infections of that group. Hence the pattern of antibody distribution determined currently in different age groups provides a serologic recapitulation of past infection with antigenic variants of influenza viruses.

Further studies of the antibody patterns of the human population with respect to variants of influenza virus may provide a clearer view of the time-honoured problem of influenza cycles and periodicity. If the suggestions observed in the present results are substantiated, the possibility exists that the times of major changes in antigenic structure can be charted or even anticipated.

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Professor C. H. Stuart-Harris: The mode of spread of influenza has recently been studied chiefly by the method of recovery of strains of virus from epidemics in different geographical areas followed by serological comparison of such viruses in the laboratory. In Europe, epidemics studied at the World Influenza Centre have sometimes shown serial spread from one country to another of the same virus strain. At other times, a particular virus strain appears to have laid dormant in one country and has only spread generally months after its original recovery from man.

An investigation was carried out at the Albany Laboratory of the New York State Department of Health last winter whereby the occurrences of influenza virus infection was detected by antibody changes in the population. Sera obtained originally for Wassermann tests were also tested against the soluble influenza A antigen in the complement-fixation test, and this was done on batches of specimens from five different areas in New York State. Although no general epidemic of influenza occurred in the State, the percentage of sera with positive fixation of complement altered during the winter in each area but at different times. Subclinical infection appeared largely responsible for these changes and it seemed likely that outbreaks of clinically detectable disease depended on the particular local conditions which may have favoured spread of infection. The importance of further study of the serological status of populations in interepidemic and pre-epidemic periods became evident during the investigation.

[*March* 19, 1954]

Social Class Variations in Mortality [*Summary*]

By W. P. D. LOGAN, M.D., Ph.D., D.P.H.

INFORMATION about the occupation of deceased persons is obtained as a routine in England and Wales at death registration, and similar information as regards the population in general is obtained at each census. At approximately decennial intervals since 1851, the General Register Office has been able to calculate occupational mortality rates for a variety of different occupations and causes of death. Less comprehensive studies have occasionally been undertaken in other countries, e.g. France, Netherlands, Scotland, Switzerland and U.S.A.

The occupations distinguished in the census and mortality tabulations for England and Wales have been grouped into a number of so-called Social Classes, assignment of occupations to these Social Classes depending upon their general social and environmental characteristics. The Social Classes are I Professional, II Intermediate, III Skilled, IV Partly Skilled, V Unskilled.

An analysis of mortality in England and Wales in 1950 in these social classes has just been completed (Registrar-General, 1954. Decennial Supplement, 1951. Occupational Mortality. Part I. London; H.M.S.O.), utilizing as denominator the 1951 Census 1% Sample tables, for men and for married women aged 20–64 and 65 and over, the married women being classified by occupation of husband. In general, married women share the same socio-economic environment as their husbands but are not subject to their specific occupational risks. Comparison of the mortality of men and of married

women may therefore distinguish mortality risks of occupational origin from those that are influenced more by socio-economic factors.

In previous studies (1921–23 and 1930–32) the mortality rates for men and for married women (all causes of death) increased progressively from Social Class I to Social Class V, but in 1950, for reasons which have not yet been determined, Social Class II (men and married women) and Social Class IV (men only) recorded unusually low mortality rates (Fig. 1; *see also* Fig. 2 in *Brit. J. Soc. Med.*[1]).

Amongst individual causes of death that were studied in the 1950 analysis steep gradients of mortality increasing from Social Class I to V were given by respiratory tuberculosis, cancer of stomach, myocardial degeneration, bronchitis, pneumonia (Fig. 3) and ulcer of stomach (Fig. 3). A mortality gradient in the opposite direction was given by leukæmia. Coronary heart disease

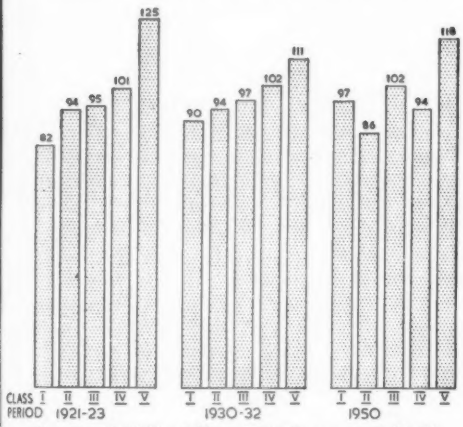

FIG. 1.—S.M.R.s (all causes) of men aged 20–64 by social class.

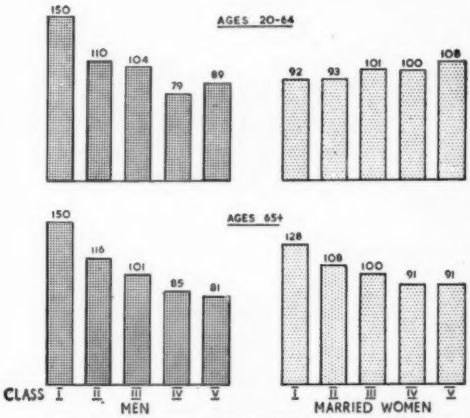

FIG. 2.—Coronary heart disease. S.M.R.s of men and married women at ages 20–64 and P.M.R.s at ages 65 and over by social class. 1950.

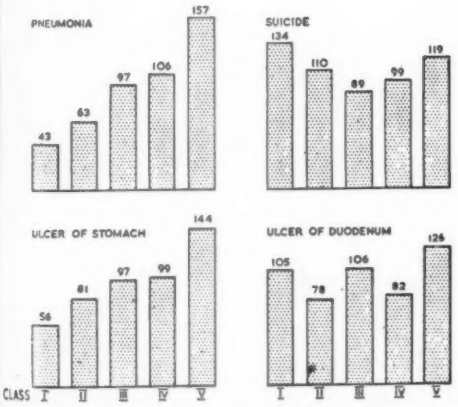

FIG. 3.—S.M.R.s of men aged 20–64 by social class. 1950.

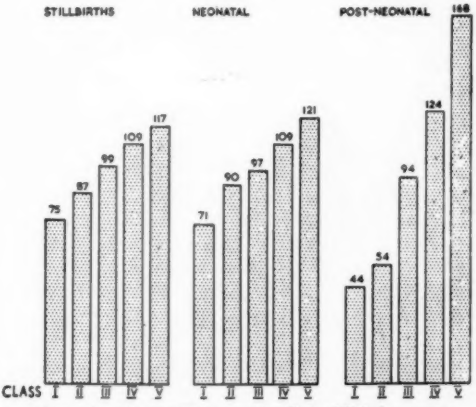

FIG. 4.—Comparative ratios of stillbirths, neonatal (under 4 weeks) and post-neonatal (4 weeks to 1 year) infant mortality by social class. 1950.

(Fig. 2) and diabetes mellitus gave high rates of mortality amongst men in Social Class I, declining to low rates in Social Class V, but amongst married women the mortality gradients were in the opposite direction. Cancer of breast gave a downward gradient, and cancer of uterine cervix an upward gradient amongst married women from Social Class I to V.

Stillbirth and infant mortality rates increased progressively from Social Class I to V, especially amongst infants dying at ages between 4 weeks and 1 year (Fig. 4). This social class gradient of infant mortality was as steep in 1950 as it was thirty years earlier.

[1]This paper will appear in full in the *British Journal of Social and Preventive Medicine*, July 1954. Figs. 1–4 are reproduced from that journal by permission of the Editor.

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Epidemiology in Modern Perspective

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EPIDEMIOLOGY has to do with the physiological state of communities and with the diseases of man as they affect populations of people. Along with clinical medicine and experimental pathology, it is one of three general methods by which progress is directed toward improved prevention and a better clinical management of disease. Its distinguishing characteristics are the study of disease as it occurs under natural conditions, and by a unit of observation which is the whole of a population of people, animals or arthropods.

Health and disease are also to be observed in hospital or consulting room, or in the experimental laboratory, where the unit of observation is one individual person, or an animal in which disease is induced experimentally. Epidemiology is in no sense a substitute either for clinical medicine or laboratory experimentation. Each method supplements the other two.

The level at which investigation is directed usually determines the method of study (Gordon, 1950). For micro-organisms and disease of cells or a tissue, the approach is by laboratory methods. Clinical study finds its main application at the level of organs and of the whole individual. The field methods of epidemiology come into play when the unit of study is a population, aided to an extent by the newer techniques of experimental epidemiology, applicable both to man and to animals. Because disease is a broad biological process, advance in understanding usually depends on use of all three methods, with sometimes one and sometimes another in ascendance.

Epidemiology has been presented elsewhere as the diagnostic discipline of public health (Gordon, 1954), the means by which mass disease is measured, evaluated and prognosis determined. The identification of the multiple factors in causation permits more specific and more effective direction of prevention and control.

In common with clinical and experimental pathology, epidemiology has investigative as well as operational objectives in analysis of mass pathology. The relative emphasis in a particular situation is determined by many factors, a principal consideration being the existing state of knowledge of the disease at hand. With adequate information available, the interest is more likely toward practical application of the epidemiological method in determining procedures for control. With indefinite or incomplete information, the stress is reasonably on the search for added knowledge. Few situations within mass disease are wholly exclusive of the one or other objective; in no disease are all problems solved and conversely most diseases give some opportunity for prevention or control. Neither are operational and investigative objectives opposed and distinct. The value of operational research, the learning in the doing, is increasingly appreciated, detracting in no way from the more material contributions to be expected from planned and intensive study. The latter interest is here the main concern.

THE EPIDEMIOLOGICAL METHOD

Mass disease will be presented as the resultant of biological forces exerted by a host—in this instance a population of people—and by the environment in which that host lives. This concept comes through an evolution of ideas (Winslow *et al.*, 1952) that dates from the time disease as a mass phenomenon first engaged the attention of man; it was first formalized in terms of medical ecology by Sheldon Dudley (1936) in a Presidential Address to this Section. With disease and injury interpreted as problems of ecology, then behaviour qualitatively and quantitatively is dependent upon the kind of balance, the state of the biological equilibrium, currently existing between a living organism and its environment. In most instances this is a state that varies from time to time and under a variety of circumstances.

Recognition of multiple factors in causation is inherent to an ecological interpretation of mass disease. The simpler complex of an opposed host and environment is enlarged to advantage by separating specific agents of disease, as a group, from the total of environment. The concept of causation is thus brought in line with modern medical attitudes in classification of disease, which is according to agent. Mass disease is thus looked upon as the interaction of a triad, the members of which are a host, with man the primary concern, an agent of disease, and lastly the intricate complex of environment. The three have a relative and varying weight in any particular situation, as do factors within groups. These require quantitation, whereby independent variables are isolated from the dependent and observed in relation to the total effect. Since ætiology as a principle applies to all disease, this pattern of ecological analysis should hold for all mass disease of human beings, whatever its origin, course and extent.

The epidemiological method is understood as the use of these principles in analysis of mass disease. It is, of course, nothing more than the scientific method applied to a particular purpose, the study of health and disease in groups of people. The basic scheme involves, in order, a definition of the nature and extent of the problem; a search for features of causation of the disease and of disease

distributions, through study in nature of affected populations; the formulation of principles for a programme of control based on demonstrated cause and directed towards limitation of death, defect and disability; and, finally, field and statistical evaluation of the results derived from control measures.

Methodology has advanced progressively. The original restricted concern with epidemics represented an emergency epidemiology, the main objective to put out the fire, often empiric in its methods and practised with or without facts. With recognition of the epidemiological case as the practical unit, that being the infected individual and his family contacts, procedure was refined. Analysis of epidemics became the accumulation of such units and their established relation one to another. Case study proved of equal usefulness in endemic circumstances, as a means for limiting incipient epidemics.

The field survey of whole populations, designed to determine prevalence of a particular disease or of total disease frequency at a particular time, eventually was added to these retrospective or proband methods, which start with a known focus of disease, with the aim to retrace lines of transmission and identify the point of origin. For the infectious diseases, the lesser frequency and moment of epidemics made this development possible; it was essential in the increasing application of epidemiological method to mass injury and to mass disease of non-infective nature, where the endemic state prevails and epidemics are still less frequent.

The more recent development may be called the field review, for it is based on incidence in contrast to prevalence. Long-term prospective observation of populations is directed toward determining origin and course of diseases of slow evolution, typically the chronic degenerative and neoplastic diseases. The cost and effort are excessive. Some such studies are designed to extend over a generation, but no other procedure gives promise of the desired information.

Epidemiological method made its greatest progress when laboratory methods were combined with field study. This may be said to have started when Robert Koch left his laboratory to study cholera in Egypt. The principle has governed epidemic practice in the communicable diseases ever since. It has proved no less essential as field activities turned to other mass disease, to diabetes and heart disease, nutritional disorders and the single persisting miasma of modern times, the "smog".

The main potential of the epidemiological method in research is in identification of causes discernible in nature and difficult to determine otherwise. The greater practical usefulness often resides in ability to recognize and describe problems of mass disease, with clinic and laboratory the places most likely to settle them; but again a return to the field for the necessary tests of usefulness and practicability of the measures there determined.

The intent now is to examine some of the events engaging the attention of field workers in epidemiology.

Communicable Disease

The summary attention here accorded the communicable diseases is by choice, arbitrarily taken and implying no failure to appreciate their relative position among epidemiological activities, now or in the past. No better reason is at hand than a wish to speak of some of the newer and less well-developed fields. A modern perspective of epidemiology recognizes that in many parts of the world, and in countries accounting for the bulk of world population, the infections have first rank, with every indication that they will so continue for a long time. In the Western world they have relatively less significance than fifty years ago. Other mass diseases have gained place, to an extent warranting partial diversion of resources toward their study and limitation.

There are many reasons for a continuing and sound concern of infectious processes. Tuberculosis and pneumonia are outstanding causes of death. Many other infections are far from control, to say nothing of the eradication that some so optimistically seek. Unsolved problems exist in plenty. Outbreaks are frequent and epidemics endure. Furthermore, the infectious diseases continue as the place "to learn the trade" of epidemiology, for more is known of their behaviour than for any other class.

Accidental Traumatic Injury

The application of preventive measures to accidental injury has lagged behind attention to disease. When "the care of the sick and injured" was enlarged to include preventive activities, injuries were neglected. The main reason was a prevailing disbelief that they were due to anything more than bad luck. That accidents have causes as definable as for many diseases and therefore are capable of practical limitation, was not recognized; as a consequence insufficient information was at hand upon which to build a programme for prevention.

The epidemiologist is readily attracted to accidents. They are acute conditions, like the communicable diseases; they even occur in epidemics and are associated with seasonal, age and sex, occupational and all manner of other behaviour manifested by mass disease (Gordon, 1949). A study was undertaken to determine by epidemiological analysis the features of accident prevention that warranted special consideration in Massachusetts (Roberts and Gordon, 1949). Accidents ranked fourth among causes of death in 1946, in both Massachusetts and in the United States, compared with sixth position in 1935. The State had a considerably lower rate than the country as a whole. Since 1923 Massachusetts has had a lower motor-vehicle accident rate than the United States generally. The record for accidental

deaths in industry was also better than average. With accidents divided into classes by place of occurrence, accidents in public places (non-motor-vehicle) accounted for a larger percentage of accidental deaths in Massachusetts than in the United States as a whole, and rates for home accidents put Massachusetts in the poorer group of States. Falls were identified as the primary factor; only one State exceeded Massachusetts in accidental deaths from falls and crushing injuries. Severe falls are a function of age and Massachusetts has an older population, in part accounting for the fact that in 1946 falls were responsible for 26% of all accidental deaths in the United States, and 44% in Massachusetts. The place to direct preventive efforts is clearly defined.

The initial proband approach through investigation of fatal accidents is now being extended through the prospective methods of field survey and field review. Total populations are the unit of observation. Both fatal and non-fatal accidents are determined, also the frequency of accident situations as well as of the events themselves. Home accidents receive the particular attention of health authorities as the social agency best suited to their control (O'Rourke, 1953), but motor-vehicle accidents also are being studied by these newer methods (Beadenkopf *et al.*, 1954).

Cancer

Cancer was an early epidemiological interest among non-infective diseases, largely through the initiative of the late Professor Greenwood (1935). Current effort is mainly toward description of characteristics of the kinds of people who do or do not develop the disease, as a means to apply such cancer control methods as exist (McKinnon, 1950), chiefly early recognition and clinical management of patients. The classic example is that of Sir Percivall Pott who observed that most of his patients with cancer of the scrotum were chimney sweeps; he made the association between soot and scrotal epithelioma. Such observations from an occupational standpoint more or less dominate epidemiological activities, but an extension of cancer epidemiology is seen in studies of geographical distribution of cancer of skin and other sites (Lombard and Potter, 1950), and a search for the causative factors involved.

Two methods illustrating a descriptive epidemiological approach (Gilliam, 1953) are indirect study of cancer through proband cases and direct measure of the disease in a population. The proband method obtains histories in retrospect on patients with cancer and a selected group of persons without cancer. The method is valuable, but fails of information about incidence in the population at risk, for patients are not always representative of the population from which they are drawn. Cancer of the lung and its association with smoking is cited in illustration. The principal epidemiological studies to date are based on study of proband cases (Doll and Hill, 1950), with demonstration that the greater percentage of patients with this form of cancer are heavier cigarette smokers and have smoked for a longer time than persons without lung cancer. The direct method is being used by the United States Public Health Service and the Veterans Administration to determine incidence of pulmonary cancer in a defined population at risk. Smoking histories are being procured from 300,000 veterans of World War I who carry government life insurance. Since approximately 4,000 to 5,000 deaths now occur each year, a reliable incidence of lung cancer presumably can be determined, with relation to history of smoking.

Definitive methodology may be applied to cancer only when a specific agent or complex has been isolated, thus providing conditions comparable to infectious agents in communicable disease. In certain instances the epidemiological approach is toward determining agents or suspect material, for example, identification of carcinogen in chromates, aniline dyes, cigarette smoke (Hueper, 1949). For the rest and indeed the bulk of cancer, this specific relationship of host, agent and environment has not been ascertained and activities continue more or less restricted to description of characteristics of population groups who do and do not develop cancer.

As cancer is studied as a mass disease, the various environmental factors to which a general population is exposed have gained significance in causal relationship, examples of which have been cited. Persons living in a sheltered environment where life is well regulated and differs from that of the general population, conceivably may have an incidence of cancer that is different. Studies are now under way to determine incidence of cancer and distribution of cancer by sites among nuns in a Catholic religious order.

Diabetes

Diabetes is a chronic disease for which no specific agent is known. The search for causative factors in this or any other mass disease is not inhibited because the agent is not in hand; if anything investigation of factors inherent in host and environment is more strongly indicated. Case study of affected individuals has established that age of onset is more frequent between 40 and 60 years, about half of the cases developing within that age group. The relative severity and atypical clinical course of diabetes in younger people compared with middle-aged and old, suggests a multiplicity of causes. Jews as a group have been historically associated with a high incidence of diabetes, but a small field survey under American conditions did not bear this out.

A search among physical characteristics of individual patients demonstrated that diabetes is certainly not distributed in random fashion between the fat and the lean. Obesity does appear to be

an important predisposing factor, at least in adults, for about one-half of diabetics in a comprehensive clinical study were at one time or another at least 20% overweight. Much remains to be done in defining the basic physiological relation between obesity and diabetes, particularly through epidemiological and laboratory procedures in supplement to existing information so largely of clinical origin.

The increased susceptibility of individuals with a family history of diabetes suggests a strong hereditary factor. This has been defined by genetic analysis and family study (Pincus and White, 1933) but the challenge of a practical prevention of clinical diabetes in individuals genetically predisposed is still open.

Recent evidence suggests that women destined to develop diabetes often suffer temporary defect of carbohydrate metabolism during pregnancy. If untreated, the condition frequently results in fœtal loss, somewhat less definitely in fœtal damage predisposing the child to diabetes. The subject is under investigation epidemiologically—and clinically—through insulin treatment of such persons during pregnancy in attempt to prevent diabetes in mother and child.

Environmental factors as a determinant of diabetes are less well defined. However, wide variations in diabetes mortality have been established among States of America; highest rates are in the north-east and lowest in the south and west. Death-rates are higher in cities than in rural areas.

One community in Massachusetts (Wilkerson and Krall, 1947) has been studied by field survey and now by field review to describe the course of diabetes in a group of people. The original survey gave a prevalence of about 1·7%, with some half of cases previously unrecognized. Characteristics of individual cases were determined. In subsequent survey, those with borderline laboratory findings at the first examination were firmly defined as a high incidence group. Currently, comparison is being made of physical characteristics of patients, of borderline cases, and of controls. The behaviour of diabetes in one community of 5,000 population should be better understood as this study is completed.

Congenital Anomalies

Probably the most striking epidemiological fact unearthed about any of the congenital defects is the relation of mongolism to advanced maternal age; this observation is also the longest established. The risk of producing a mongoloid child increases about 50 fold, from around 0·3 per 1,000 pregnancies at age 20–30 to around 15 in the fifth decade. A beginning control of this life-long defect of the central nervous system is to be expected only when these epidemiological facts can be interpreted, expanded and given meaning. What are the factors associated with ageing of mothers which synchronize to maim the differentiating brain, heart, eyes and little finger? Are they endocrine, gynæcological, systemic or combined? The answers to such questions which are biological, not statistical, are the crux of the problem of control. Geographical studies of mongolism have never been done on anything more than a tentative and inadequate basis, nor have secular occurrences or changes in maternal age distribution in different countries and at differing times been plotted, partly because mongolism occurs so rarely and partly because epidemiological methodology has barely begun to be applied to congenital defects.

The indications for applying the epidemiological method to problems of congenital defect were inherent in the original observations of Jonathan Hutchinson on the congenital stigmas of syphilis; an actual urgency to do this was revealed when Gregg demonstrated the teratogenic effect of rubella in pregnancy. The determining importance of conceptual age of the embryo and fœtus emphasized in 1947 (Ingalls and Gordon, 1947) may be shown to operate throughout prenatal life and at the earliest moments after premature birth. The condition of retrolental fibroplasia (Ingalls and Purshottam, 1954) further exemplifies how epidemiological methodology may be fruitfully applied to the solution of an unknown disease, with the differentiating retina the susceptible tissue and the newborn fœtus the susceptible host.

Epidemiological studies are currently in course at a number of centres, designed to analyse the relation of mongolism to maternal ageing. The relation itself suggests that chronic rather than acute processes are involved. A definite excess of chronic conditions—manifestations of heart and kidney disease, gynæcological disturbances and anæmias in older mothers—has been observed in 50 pregnancies productive of mongoloid babies compared with 50 other pregnancies selected as controls, mothers being matched for age. The two groups are being compared according to various biological attributes of the mother, for mongolism is interpreted as due to one of several non-specific stresses active in the first trimester of pregnancy.

Nutrition

The field approach to disease has perhaps seen higher development in nutrition than in any other mass disease excluding infections. The work of Goldberger *et al.* on pellagra in 1916 stands with that of Snow, Budd and Panum among classical field studies. I would think that James Lind (1753) had a pretty good idea of the epidemiological method although he scarcely would have called it by that name. Wading through the wilderness of conflicting theories and the mud of the mill villages of South Carolina, Goldberger found his data in the homes of persons affected, and proved conclusively that the disease was due to defect in the diet consumed. His ideas enlarged to include the

potentiality of a protein as well as a vitamin deficiency, both currently recognized as interlocking factors of causation of pellagrous disease.

The American concern with obesity is a never-ending source of bewilderment to foreign students coming to Boston. The problem is not exactly world-wide, but the increasing interest in chronic degenerative diseases establishes important relationships. These have been pursued in a number of directions by clinical and laboratory investigations. My colleague Jean Mayer (1953) has recently brought them together in a summary of the community problem. He sets down three sets of factors relating to the triad of host, agent and environment, which in obesity include heredity, bodily or mental injury, and various features of the ecological environment such as food supply, temperature, exercise and physical activity, and socio-cultural mores. An interaction of multiple causative factors is seen in explanation of the broader community reaction.

A casual acquaintance with Asia and Africa suffices to sharpen appreciation of protein malnutrition as a global problem. Within the past decade a condition known as kwashiorkor has emerged into world medical literature, largely as the result of field surveys by Brock and Autret (1952) for the World Health Organization and the Food and Agriculture Organization of the United Nations. Kwashiorkor has been established as the extreme degree of protein malnutrition in a biological gradient (Gordon, 1950) that extends from the mildest forms with little or no clinical manifestations through intermediates with known and as yet unknown features. Protein deficiency is thus being fitted into a pattern analogous to that of poliomyelitis and diphtheria, far-reaching in the people it involves and with all manner of variation in the impress upon populations of people. The community studies are in the best tradition of field epidemiology.

Other Non-Infectious Mass Disease

The continued cataloguing of contemporary interests in field study would prove tiresome were it not also beyond this brief summary. Some few are named: (1) The Framingham Heart Study (Dawber *et al.*, 1951), now four years under way in a projected investigation to extend twenty years, an outstanding example of those long-continued observations which appear the necessary approach to mass disease of degenerative and neoplastic nature; also the studies of Morris *et al.* (1953) which we follow in America with much interest; (2) Smog, the single persisting miasma of modern times (McDonald *et al.*, 1951), of increasing consideration with greater industrialization, in your country (Report, 1953), and in mine (Schrenk *et al.*, 1949); (3) Chemical intoxications, not alone in industry but in general populations, so well brought out by Baltimore observations on lead poisoning (Williams *et al.*, 1933) but dating back to the Devonshire colic (Baker, 1767); (4) Dental abnormalities in which Trendley Dean (1938) has taken the lead, with community studies (Ast *et al.*, 1950; Schlesinger *et al.*, 1950) now centring particularly on fluoridation in relation to dental caries.

The emphasis, thus far, has been on planned investigation by groups of investigators directed toward acquiring fundamental knowledge in selected fields, always of course with an aim of ultimate use in a better programme of prevention. I do not forget those contributions which form so large a part of knowledge of mass disease, the contributions that come from what is now more circumspectly known as operational research, the things that are learned in the course of the doing, that come from the careful recording of events as they develop during active practice of public health; I refer to the Pickles (1939) kind of epidemiology. My single feeling is that the scope of these observations should now be broadened. Unsolved problems of communicable disease are on every hand; infections continue as the diseases with which health authorities are most familiar. They rightly interpret them as the primary concern. However, altered conditions of incidence and administrative responsibility make possible an attention to other community diseases of recognized importance and relatively increasing concern. I suggest the epidemiology of accidents and of mental disorder as worthy endeavours of official health agencies. I turn now to general rather than specific fields of interest in investigative epidemiology.

Geographical Pathology

For the greater part of the nineteenth century the dominant approach to epidemiology was through geographical and historical pathology. Its potentiality was greatly enlarged through the field method of study that came into play in the last half of the century. The full value of its contribution might have been attained with the coming of microbiological methods, except that the study of disease was then to a large extent lost to view in the newer enthusiasm for the study of the agents that cause them.

With the twentieth century, medical geography came close to being a lost art. World War II demonstrated the usefulness and the necessity of this sort of information. An Atlas of Disease (Zeiss, 1942–44) was prepared in Germany under military auspices and a comprehensive geography of disease and sanitation (Simmons *et al.*, 1944, 1951) in the United States. The stimulus continues (Light, 1944).

The development of facilities for scientific investigation and increasing numbers of trained men in tropical countries have produced an imposing information on specific features of mass disease under these conditions. Problems indigenous to the tropics become a matter of general investigation

as the world tends to become a single epidemiological universe, with military and political considerations adding incentive. Other than for the little book of Edge (1944), I know of no attempt to bring these things together in a geographical pathology of the tropics. What was known in the days of Hirsch (1886) and others, centred largely in Western Europe and to an extent in America. The epidemiology of helminth and protozoal diseases is still too much restricted to life cycles of parasites and broad regional distributions, despite the example of malaria which is unsurpassed in any part of epidemiology. The problem of accidents in less well-developed countries is unexplored; what is known of cancer as a community disease is clearly not transferable from the Western world, on which it is based, to countries of the tropics.

The variations in mass disease occasioned by arctic or cold weather environment are scarcely beyond the speculative level. These regions become important with expanding commerce, the need for more land, especially mineral deposits, and militarily. The behaviour of the simplest diseases is unknown, the relative place of contact and vehicle transmission of intestinal infections for example. Because the tropics are the foreign environment best known to workers in temperate climates, the tendency is to transfer that knowledge unwarrantedly to this other strange environment. Such work as has been done is largely under simulated conditions in the laboratory. The call is for field work—again the study of disease in its natural environment. Pioneering in epidemiology is still possible.

Experimental Epidemiology

An attempt to define the general laws and principles of mass disease by experimental means was a logical step in the transition that science experienced generally, as it progressed from descriptive and comparative method to experiment. The ground work was by Greenwood *et al.* (1936), Webster (1946), by Schneider (1946) and others in America and, most recently, by Fenner (1948*a*, *b*) in Australia. Burnet's observations (1952) on myxomatosis of rabbits revive the methods of Theobald Smith (1934), in studying natural disease in animal populations. Dingle and his group (Badger *et al.*, 1953) in Cleveland have accomplished an innovation in methodology through substituting human populations for animal. The method of natural entrance of acute infection into the herd is determined through continued observation. Its course, behaviour and subsequent result are measured by periodic examinations.

Epidemiology of Health

In interpreting the problems of populations having to do with soundness of body and mind, emphasis may well be placed on the fact that both health and disease are resultants of an ecological process. It is therefore singular that in applying epidemiological methods to analysis of practical problems, the events or things selected as the basis for study have been so regularly disease or injury. This view has held throughout the history of epidemiology, despite the increased scope that epidemiological interests have taken. The word epidemiology, signifying "upon the people" and referring to occurrences that affect man in the collective sense, can reasonably include events that are either favourable or unfavourable, hence both health and disease. The concept of an epidemiology of health has been set forth in a recent publication of the New York Academy of Medicine (Gordon, 1953), with illustrations of study and practice under a variety of conditions.

The knowledge essential to practical use of an epidemiology of health may well be forthcoming in the many studies of whole populations now so much a feature of epidemiological practice (Perrott *et al.*, 1939). Investigation has turned also to several specific items such as maternal age (MacMahon and Gordon, 1953) and premature birth (Corsa *et al.*, 1952) as determinants of community health. Still more recently, controlled field studies have been initiated on a ten-year basis, to determine by direct observation the potentialities of controlling overpopulation in a specific area (Gordon *et al.*, 1954). A further objective is to determine what effect control may have on the economic and social status of a population, if successfully obtained, a consideration almost universally assumed to be of favourable nature but never proved; much less, quantitated under controlled conditions. Cheap, simple, safe and reversible methods of contraception are known. Clinical trial has been adequate to demonstrate usefulness. What may be expected in the way of results when these methods are applied to whole populations has yet to be determined. The critical issues are biological efficiency of the method when used outside the ordered conditions of laboratory and modern medical clinic, and secondly, acceptability to the particular population. The study is epidemiology in a most attractive aspect, for causative factors are both biological and social, methods of study involve both sets of disciplines, and measurements are in similar dual terms. With judgment based on exigency and the welfare of a total world, the problem of population dynamics is surpassed by few.

Summary and Conclusions

As the diagnostic discipline of public health, epidemiology finds increasing use in the definition of health problems, in determining principles to guide programmes for control and in evaluation of accomplishment. The promise of a more scientific and a more statesmanlike public health has a close relationship with operational epidemiology. With all mass disease and mass injury understood as the resultant of ecological forces within a prescribed universe, epidemiology has extended its activities

from communicable processes, to find application in organic diseases of non-infective origin, in mental disorder and in the problems associated with trauma. Through planned investigation and prospective observations as additions to the usual comparative and analytical approach to past events, field studies on human populations have the potentiality of an improved instrument for research.

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Section of Odontology

President—ARTHUR BULLEID, F.D.S., L.R.C.P., M.R.C.S.

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The Muscles of the Mandible

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IT is usually very difficult, and in many cases impossible, actually to prove what is the action of a muscle. Although the writers of textbooks of anatomy do not hesitate to state categorically the action of every muscle they describe, they seldom quote any evidence. It does not follow that an eminent anatomist who is very exact in his account of structure is equally correct in his interpretation of function. If errors of factual description of structure creep into a textbook (and rare is the book that is free of them), the author's accounts of function will be correspondingly less convincing. So it would appear that the answer to the question, "What is the action of this muscle?", is not necessarily to be found by consulting an otherwise authoritative textbook of anatomy. In many cases, indeed, the answer is scarcely worth finding, since muscles do not contract alone, but usually in a pattern in harmony with synergic members of their own group and always either against their opponents, which "pay out rope," or by themselves "paying out rope" against gravity.

In some cases, however, it is very important to have an accurate knowledge of muscle action. To the ophthalmologist, for example, the action of each muscle that moves the eyeball is of signal concern, since he must assess the implications of strabismus and diplopia. Likewise to the orthodontist the actions of the muscles that move the mandible must be of equal interest, since the contours of growing bone seem in large measure to be adjusted to the forces of muscle pull. Rix, Ballard, Gwynne Evans and Tulley in this country are among many who have emphasized the importance they place on the role of the muscles that are used in swallowing, and Brodie in the U.S.A. has advanced a conjecture about the supposed role of these same muscles in preserving the balance of the skull on the spine. If orthodontic study is to advance our knowledge of the relationship between muscle action and bone form, it is essential to know very precisely what are the normal actions of the muscles being studied.

The present writer has been singularly dissatisfied with several of the orthodox and accepted statements concerning the actions of muscles in and about the floor of the mouth, and in particular with the usually accepted accounts of what muscles are responsible for opening the mouth. This paper is concerned with an attempt to assess the reasons for the presence and contours of certain muscles, and especially to understand the essential arrangements of the muscle groups involved in natural movements of the mandible, tongue, larynx and face.

The problem of the action of a muscle can be attacked from several directions, some of which are listed here:

(1) Speculative reasoning and deduction, or "armchair anatomy" if the reader prefers that term. This is a dangerous method, for anatomists are notoriously poor at mechanics; but it remains the most commonly used approach.

(2) Feeling the muscle in contraction in the living. This has only a limited application, for many muscles are not palpable.

(3) Pulling on the muscle or its tendon in the dead. This is often impossible (e.g. in flat sheets of muscle like the mylohyoid) and it is usually unconvincing, since the background of tone in opponents is absent and synergists are out of action.

(4) The effects of paralysis. This may give a "negative print" of muscle action; but in a host of cases failure results from the fact that other muscles are employed to produce "trick" movements, and the paralysis is masked.

(5) Stimulation of a muscle or its motor nerve, exposed or not, with or without anæsthesia. This is usually unconvincing, since the background of tone in opponents is absent or diminished under anæsthesia, and synergists are likewise out of action.

(6) Action currents in the muscle during movement:

(a) by surface electrodes, a somewhat rough and ready method, or

(b) by needle electrodes. This is possibly the most accurate method available, although spread of current from near-by muscles cannot always be ruled out. Moreover, action potentials recorded from a muscle during a given movement do not prove that the muscle is responsible *as a prime mover* for that movement. The muscle may be contracting synergically.

(7) Comparative studies. An assessment of the form and function of a similar muscle or group of muscles in a comprehensive series of other vertebrates will often, though not always, throw light on the action in man.

Study of the results obtained from the above methods of approach shows that none gives a wholly objective answer, for analytic and synthetic reasoning are necessary to assess the results of whatever method is used. It becomes doubtful if, after all, the best method is not, in fact, to "work it out for oneself."

Realizing the pitfalls, and endeavouring to overlook no significant fact, the writer therefore proposes to reason out the actions of the muscles in the groups under consideration. Certain fundamentals will be admitted to be true by everyone. If, for example, it can be shown that a certain movement causes the attached ends of a muscle to be pulled farther apart, for the muscle to be elongated, that particular movement cannot possibly be the action of that particular muscle. On the other hand, if a movement approximates the attached ends of a muscle it is reasonable to assume that the action of the muscle *may be* to produce that movement, if not as the prime mover, at least to take part as an assistant in the movement.

The plan of the argument is based on the following simple premises:

(1) The skull is movable on the spine in all positions of the latter.

(2) The mandible is movable on the skull in all positions of the latter.

(3) The floor of the mouth is movable on the mandible in all positions of the latter.

(4) *There is a separate set of muscles provided for each of these three movements,* and confusion has arisen from failure to place certain muscles in their correct functional group.

The Movements of the Skull

The skull rests on the top of the spine like a ball on a flagpole. Unsupported it would fall forward, for its centre of gravity lies anterior to the occipital condyles. Guy ropes stabilize and, when required, move it. They are the extensor muscles at the back of the neck, opposed by the flexors, which are the prevertebral muscles and the sternomastoids. The fact that the flagpole is a flexible, segmented structure does not invalidate the above statement.

There is a set of balanced extensor and flexor muscles to stabilize and move the cervical spine independently of movements of the skull. It should be noticed that muscles moving the skull will also of necessity cause secondarily a bending of the cervical spine unless such effects are neutralized by the opposing spinal muscles.

Extension of the head is brought about by contraction of the long extensor muscles at the back of the neck, most especially by semispinalis capitis, which muscle if unopposed must also extend the cervical spine. The rectus capitis posterior minor must also have an extensor action of occiput on atlas and it is difficult to picture the reason for the existence of such a minor muscle. No doubt it is a factor in stabilizing the atlanto-occipital joint. It is just possible that it may also be responsible for the slight extension of skull on atlas that accompanies opening of the mouth. It has no effect on the cervical spine, arising as it does from the atlas; if skull extension is produced by any other muscle the secondary extension of the cervical spine has to be balanced by the opposing flexors, a necessity that does not arise in the case of rectus capitis posterior minor. It would certainly seem to be admirably placed to produce that slight and uncomplicated head extension which accompanies depression of the mandible. The muscle is too deep for accurate palpation. If, however, I press deeply into my suboccipital region I fancy I feel a deep resistance when I open my mouth. This may be imaginary and I do not press the point. In any case the matter is of purely academic interest, and need not concern the orthodontist. The rectus capitis posterior major is badly named; it is not erect, but lies obliquely, and is primarily a head rotator.

The opposing flexor muscles consist of the prevertebral group, must less massive than the extensors because they are assisted by the sternomastoids and, in the erect position of the body, by gravity. Longus capitis, formerly named rectus capitis anterior major, is the only long skull flexor in the prevertebral group. The rectus capitis anterior (minor) connects skull and atlas, and would seem to be the opponent of rectus capitis posterior minor. Longus cervicis (colli) and scalenus anterior are flexors of the cervical spine.

For the true opponents of the extensors we must go farther forward, to the sternomastoids. Here are two very powerful flexors. Much ink has been spent discussing the action of these muscles on the skull. Do they flex or extend it? That they pull the cervical spine forward into

flexion nobody doubts, but many will have it that they extend the atlanto-occipital joint. It is almost certain that the posterior fibres of the muscles are, indeed, extensors of the skull at the atlanto-occipital joint, at the same time having the secondary effect of flexion of the cervical spine.

Examination of the skeleton, of a dissection, and of the living neck all confirm this idea, for these posterior fibres of the muscle pass up posterior to the atlanto-occipital joint. But the most anterior fibres of sternomastoid, those which arise from the sternal head and pass to the projecting tip of the mastoid process, are flexors of the atlanto-occipital joint.

Lie face upwards on the floor, finger and thumb placed lightly over the *anterior* borders of the sternomastoids. Flex the head against gravity and feel the powerful contraction of the sternomastoids, and do not doubt that they are flexors of the skull as well as of the neck. During this flexion of the head against gravity palpation of the masseters will show that they remain quite relaxed.

The muscles of mastication play no part in head movements or in the maintenance of head posture, and the American orthodontist, Brodie, is mistaken in his assumption that they do so.

Brodie correctly extends the human head with the post-vertebral neck muscles, but he incorrectly opposes these anti-gravity extensor muscles with a chain of muscles consisting of the muscles of mastication (temporalis and masseter, which join the mandible to the skull), the suprahyoid muscles that join the hyoid to the mandible (Brodie lumps them all together as though they all share a common action!) and the infrahyoid muscles that join the hyoid bone to the pectoral girdle, and he talks of the reciprocal inhibition of the infrahyoid muscles that accompanies contraction of the temporal muscle and elevation of the hyoid bone in swallowing.

Brodie has three links in his chain of skull-flexing muscles. If one of these muscle links is inhibited (according to the principle of reciprocal inhibition discovered by the "English physiologist Sherrington") surely the whole chain of muscles is correspondingly weakened! It would seem that the head should jerk back willy-nilly into extension every time we swallow. But Brodie, not content with ignoring the flexor muscles of the skull, stops short in attaching his own chain of imaginary skull-flexing muscles to the pectoral girdle. For the pectoral girdle is not fixed. It moves up and down with respiration, with upper limb movements, with abdominal contractions such as defæcation, and so on.

If Brodie wants to oppose his skull extensors with the three muscular links in a chain from skull to pectoral girdle he must surely hold his pectoral girdle down. This is done by the rectus abdominis (far from the usual field of investigation of the orthodontist, though it is an attractive thought that an umbilical hernia might be the cause of malocclusion!). But the rectus abdominis can only hold down the thoracic cage and pectoral girdle if the pelvis is fixed. The rectus abdominis is attached to the body of the pubic bone, and the pubic bone can be held down only by those muscles that connect it with the femur—adductor longus, pectineus and the rest. How can Brodie say that the infrahyoid muscles relax to allow the hyoid bone to ascend in swallowing? It is just as logical to argue that pectineus relaxes for the same purpose. Let the believers in the Brodie conjecture look to the pectinei and adductores longi while they are assessing the causes of malocclusion in their patients, and ponder whether this *reductio ad absurdum* does not indicate that Brodie's premises are quite untenable.

Extension and flexion of head and of cervical spine having been accounted for by the paired semispinales capitis and sternomastoids, with other weaker muscles, it only remains to say that acting singly these muscles are rotators or abductors of the spine and head.

These movements of the skull and of the neck are of little interest to the orthodontist and are mentioned here only to exclude them from the arguments about the mandible and the floor of the mouth, *each of which is entirely independent of head position and movement and is served by a separate musculature.*

The Movements of the Mandible

The resting position of the mandible is one of slight separation of the teeth, by a few millimetres at the incisors. From the resting position or from occlusion the hinge movement of opening is accompanied by a forward movement of the mandibular head from the glenoid fossa down the slope towards the articular eminence of the zygoma. Thus two simultaneous movements take place in the mandibular joint; the hinge movement occurs at the lower, the gliding movement at the upper compartment of the joint. The lateral pterygoid is rightly allotted the function of producing the forward movement, but a considerable collection of muscles are considered responsible for the hinge movement. Those usually named are the mylohyoid, geniohyoid and the anterior belly of digastric, to which are often added the infrahyoid muscles, gravity and even the innocent platysma!

The truth of the matter is that consideration of the movements in the mandibular joint, and the muscles responsible for each movement, complicates what is, in fact, a perfectly simple movement, namely a rotation of the mandible about an axis that passes very nearly through

the mandibular foramina. Viewed from the side the mandible rotates like a ship's steering wheel, one spoke of which touches the temporal bone for stability in all positions, making an articulation whose movements are necessarily complex, and study of which blurs the essential simplicity of the picture of mandibular movements. Rotation of a steering wheel is produced by a tangential pull on one or more spokes, and this is precisely what happens to the rotating mandible. The longer the spoke the greater is the moment of rotational pull. The farther the muscle is inserted from the axis of rotation of the mandible the more efficient it will be as an opener of the mouth provided its pull is in the right direction and that it can be shown to be shortened when the mouth is open.

What muscles open the mouth?—Picturing the mandible as a ship's steering wheel, it is evident that its uppermost spoke is pulled forward by contraction of the lateral pterygoid. The analogy of the steering wheel fails in one important respect, namely that there is no fixed mechanical axle around which the jaw rotates. Pulling forward the uppermost spoke of a steering wheel produces rotation about its own axle; pulling forward the mandibular condyle produces no rotation, but only protrusion of the mandible as a whole. The mandibular "wheel" is stabilized, not upon an axle, but by its uppermost spoke, the condyle, being held by the ligaments of the mandibular joint; and it is evident that to produce rotation it is necessary to pull in an opposite direction on a "spoke" more or less diametrically opposite. There is a powerful muscle that is admirably placed to perform this very movement, namely the digastric muscle (Fig. 1).

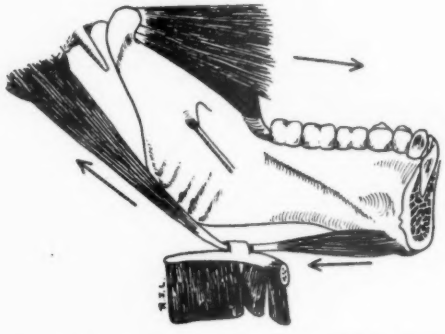

FIG. 1.—The mandible is opened by the rotational pull of the lateral pterygoid and the digastric muscles. The digastric slides freely across the hyoid bone.

The digastric muscle.—As the mandible is moved from the closed to the fully open position the chin not only falls lower but moves backwards. What muscle pulls the chin backwards if not the digastric? The common statement that the anterior belly of the digastric is responsible seems to the writer to be very naïve. What would happen if the posterior belly remained relaxed? Surely the anterior belly would simply pull forward and elongate the relaxed fibres of the posterior belly, and no mandibular movement could possibly result. How could the sterno-mastoid or any other muscle produce movement if its upper half contracted while its lower half remained relaxed? Both bellies of the digastric must obviously contract with equal force (if not with equal shortening) if any movement is to result. The fact that the two bellies have different nerve supplies has no bearing on this, since the motor nuclei of the pons are connected by longitudinal fibres, as are all the motor nuclei of the brain-stem and spinal cord.

The two bellies of the digastric do not lie in line with each other. Shortening of the contracting muscle must tend to straighten it, and thus elevation of the hyoid bone would occur before any mandibular movement could result. But the upward movement of the hyoid bone is prevented by the action of the infrahyoid muscles contracting synergically. This is the most important action of the infrahyoid muscles. To imagine that the primary reason for the existence of the human digastric is to elevate the hyoid bone is to lose sight of two facts. The first is that the muscle is placed very badly for the mechanics of such a movement and the second is that the hyoid has its own proper elevators, which will be discussed later.

In many mammals the digastric has no connexion with the hyoid bone, but passes from the base of the skull directly into the lower border of the mandible. It is the depressor of the mandible. The tethering of the central tendon in man by a fibrous sling at the lesser cornu of the hyoid bone is a refinement that possesses the great advantage of altering the direction

of pull of the anterior belly on the mandible, greatly increasing the moment of rotational pull on that bone.

The mylohyoid muscle.—The floor of the mouth is a muscular diaphragm which forms a highly mobile base for support of the tongue. This is the function of the mylohyoid, and in ordinary action *the muscle has nothing whatever to do with opening the mouth.* The action of the mylohyoid is considered later.

The geniohyoid muscle.—The action of this muscle, too, is considered later. While realizing that it contracts *pari passu* with the anterior belly of the digastric, it is submitted that this muscle acts with others to fix the position of the hyoid bone and therefore of the floor of the mouth, and it is not there primarily as a rotator of the mandible.

The essential difference between the anterior belly of the digastric (Fig. 1) and the geniohyoid muscle (Fig. 2) is that the tendon of the digastric runs freely through a pulley; the powerful digastric muscle depresses the chin by pulling from the mastoid process, without any effect on the antero-posterior position of the hyoid bone. The digastric asks only that the hyoid bone be held down, and there is a strong group of infrahyoid muscles to do that. Geniohyoid can only depress the chin if the hyoid bone is held back, and there is only a relatively feeble stylohyoid muscle to do that.

Gravity.—In the erect position it is apparent that depression of the chin is assisted to a slight extent by gravity, but it is wrong to suggest that gravity is of any real importance in the picture of mandibular movements. Several reasons support this view:

(1) Opening the mouth is just as easy when lying upside down. Watch a newborn child crying while held upside down by its legs to drain mucus or liquor amnii from its throat.

(2) A nursling lying with its face pressed sideways against a soft breast, its own light mandible supported in a firm mass of muscle and fat, could not open its mouth as briskly and quickly and widely as it does when sucking were gravity a relevant factor.

(3) The normal rapid chewing movements of the adult at his repast are too swift and of too great an amplitude to be due to gravity.

(4) Watch anyone speaking. The mandibular opening is, again, much too rapid and too well controlled to be due to gravity.

(5) Allow the muscles of mastication to relax completely and the jaw to droop. The mouth is only half open; to take a good bite from an apple a conscious mouth opening effort has to be made.

(6) What separates the teeth when they are stuck together by chewing gum, toffee or like substance? Gravity?

Platysma.—This muscle can be seen in action in an exhausted runner nearing the finishing post. While other muscles, such as risorius and the depressors of the lower lip open the lips, the platysma is an accessory opener of the jaw. But in normal movements of the mandible it is never in action. It can be called an accessory muscle of mouth opening in the same way as the sternomastoid, for example, can be on occasion an accessory muscle of respiration, but neither action has a place in the picture of *normal* movements.

Accessory muscles of mandibular depression.—All over the body we see each movement served by its own set of muscles, but in many situations it happens that additional muscles, not normally concerned in the movement, can be called upon to act as accessory muscles. This is well illustrated in the case of the respiratory excursions of the ribs. Normal tranquil respiration is carried out by the intercostals and diaphragm alone; forced breathing is performed by many other muscles not normally concerned at all in the respiratory act. For example, the scalenes, sternomastoid, pectoral muscles and serratus anterior act as accessory muscles of inspiration in dyspnœa, while rectus abdominis and the abdominal flank muscles act as accessory muscles of expiration in coughing, sneezing, asthma, &c. In the same way, under *abnormal* conditions, accessory muscles can be used to assist the lateral pterygoids and digastrics to rotate the jaw into the open position; such muscles are the geniohyoid, those few fibres of mylohyoid which are attached to the hyoid bone together with the depressors of the hyoid bone, and even the platysma.

In passing one might remark that the openers of the lips, when all contracting together and simultaneously with contraction of the orbicularis oris, can as accessory muscles assist temporals, medial pterygoids and masseters to close the mandible by stretching as a sheet of contracted muscle between upper and lower jaws. But it would cause much confusion to place the levators and depressors of the lips in a list of muscles supposed normally to close the mouth.

Closing the mouth.—The writer agrees with the conventional accounts of the muscles responsible for closing the mouth. Retraction of the mandibular condyle is brought about by the posterior fibres of the temporalis and the mandible is rotated into occlusion by the rest

of temporalis, the masseter and the medial pterygoid. The temporalis, masseter and medial pterygoid have nothing whatever to do with the head poise. The head is held in balance by its own musculature, which has already been discussed.

Lateral movements of the mandible.—It has already been stated that the mandible rotates around an axis that passes through each ramus near the mandibular foramen. But this axis does not remain fixed in relation to the skull. The mandible does not close like the hinge of a rat trap, but approaches the maxilla with a sideways movement that is essential to both cutting and grinding. The muscle balance which is responsible for stabilizing and moving the mandible is a mechanism that can be adjusted to allow asymmetry in the amount of forward movement of the condyles. Such asymmetry allows the mandible to be slewed to one or other side. The pterygoids, both lateral and medial, are essentially the muscles responsible for these lateral mandibular movements, the lateral pterygoid in opening and the medial pterygoid in closing the jaw. The grinding motion of the molars is a combination of lateral with some antero-posterior movement of the mandible, and this is produced by the pterygoid muscles of right and left sides contracting alternately while upward pressure of the mandible is maintained by temporals and masseters.

Movements of the Floor of the Mouth

The floor of the mouth, the mylohyoid, is a muscular "diaphragm" whose resemblance to the pelvic floor is very striking. In each case a flat sheet of muscle slopes downwards from a curved bone into a mid-line raphe. When the muscle is relaxed the gutter between the two sides is at its deepest; contraction of the muscle raises the floor and makes the gutter more shallow. The main difference lies in the relative fixity of the pelvic floor. The mid-line raphe of the pelvic floor is attached posteriorly to the tip of the coccyx and is there fixed in relation to the pelvic wall. The mid-line raphe of the oral floor is attached posteriorly to the hyoid bone and is thus very mobile in relation to the mandible. The hyoid bone can be moved forwards and backwards, upwards and downwards, and rotated around a transverse axis. Put in other words, the floor of the mouth can be lengthened or shortened antero-posteriorly, and can also be elevated or depressed with relation to the mandible. Such mobility confers a like mobility on the tongue, which possesses great intrinsic mobility of its own. The mobility of the floor of the mouth greatly increases the range of excursion of all the tongue movements. The position of the hyoid bone along the antero-posterior range is determined by the relative lengths of the geniohyoid and stylohyoid muscles (Fig. 2). Each of these muscles has a secondary

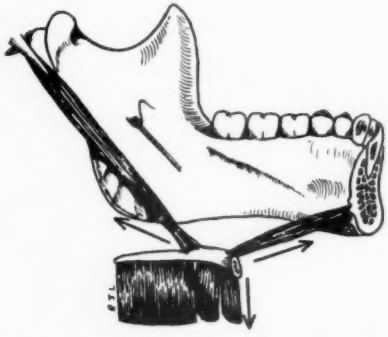

Fig. 2.—The antero-posterior position of the hyoid bone (i.e. the length of the floor of the mouth) is determined by the stylohyoid and geniohyoid muscles, both of which are fixed to the hyoid bone.

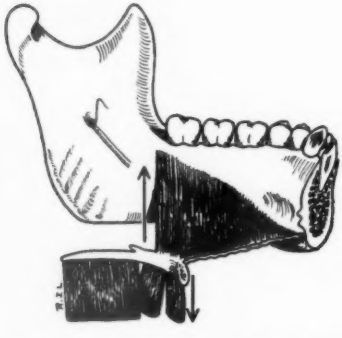

Fig. 3.—The vertical level of the hyoid bone is determined by the mylohyoid and infrahyoid muscles.

elevating effect similar to that of the digastric. This is counterbalanced by the depressing effect of the infrahyoid strap muscles. Sternohyoid and omohyoid act directly as depressors, while sternothyroid acts directly via the thyrohyoid muscle.

The vertical position of the hyoid bone is determined by the upward pull of the posterior fibres of the mylohyoid muscle, balanced by the opposing infrahyoid muscles (Fig. 3).

The principal elevator of the hyoid bone is the mylohyoid muscle. In other words, the floor of the mouth is its own elevator. The mylohyoid is a sling lying under the tongue, and when it contracts it lifts the tongue upwards. Tongue movements are essential to normal speech and mastication, but they accompany also a simple uncomplicated opening of the

jaw. It is almost impossible to keep the tongue in the same position relative to the mandible while the mouth is opened; the tongue is protruded, retracted, heaped up or flattened according to the purpose required, and depending largely on whether nose or mouth breathing occurs.

The hyoid bone is moved by its own muscles so that it takes up its appropriate position in all positions of the mandible; the muscles that so move it are not the primary movers of the mandible. Depression of the mandible is usually accompanied by contraction of the mylohyoid, but the contracting mylohyoid does not *produce* the mandibular movement. In the same way, depression of the mandible is usually accompanied by opening of the lips, but it would be absurd to argue that the dilator muscles that open the lips produce mandibular movement, even though they contract simultaneously.

Thus it is evident that the hyoid bone is movable in all directions from its position of rest, and in practice it turns out that hyoid movement in respect to the mandible accompanies the slightest movement of the tongue. In speaking and chewing the hyoid is never still. Its greatest excursion takes place during swallowing. Here the floor of the mouth is elevated to its highest, reducing the volume of the mouth to the minimum, so that the tongue must go back, propelling the bolus into a pharynx drawn up and open, waiting to receive it. The posterior part of the tongue is not only pushed back passively by the elevation of the floor of the mouth, but is pulled back actively by the stylo-glossus muscle. Thus is accomplished the first or voluntary stage of swallowing.

The Role of the Tongue in Swallowing

When the mylohyoid contracts the tongue is not pushed upwards as an inert mass. By the active contraction of its own musculature the tongue adapts its shape to the requirements of the action being performed. When the tongue is in fact inert, speech, mastication and swallowing are all very adversely affected. This is seen in idiots, or in cases of bulbar palsy where the hypoglossal nucleus is involved. The assumption that the tongue is relatively passive during the mylohyoid stage of swallowing seems to the writer to be a fallacy in the present-day assessment of the effects of swallowing and tongue movements in the production of malocclusion.

During continuous drinking of liquid from a vessel, swallowing is performed with teeth and lips apart, yet no liquid is squirted back into the vessel. This action of the tongue, in which complete control of the liquid bolus is obtained without assistance from the teeth, is surely the ideal swallow. It is the usual swallow in many native races, particularly in those who masticate with the lips parted, and it is referred to below.

It should be noted that when contraction of the mylohyoid forces the tongue up against the palate there is necessarily an increase of pressure within the mouth. This increased pressure can only be sustained if the mandible is held up. Thus the elevators of the mandible contract with appropriate force not only during mastication of a soft bolus between the tongue and the hard palate, but also while the tongue is pressed upwards during the mylohyoid phase of swallowing. Brodie draws attention to this simple fact, and to the reciprocal relaxation of the infrahyoid muscles; but Brodie's mistake lies in ignoring the flexor muscles of the cervical spine and of the skull, and in assuming that the swallowing muscles are muscles of head posture.

Movements of the Larynx

It is of great importance to the protection of the human larynx that its inlet be simultaneously updrawn below the protective shelf of the posteriorly bulging tongue as the bolus passes into the pharynx. This explains the attachment of the larynx to the floor of the mouth, via the hyoid bone and the thyrohyoid muscle; as the floor of the mouth ascends the larynx ascends with it. In addition the larynx has its own proper elevators, those muscles wrongly styled "pharyngeus", the stylo-pharyngeus, salpingo-pharyngeus and palato-pharyngeus, each of which has most of its fibres inserted into the thyroid cartilage and only a few into the side wall of the pharynx. They, as well as the thyrohyoid, draw the larynx up in swallowing.

Movements of the Face

The muscles supplied by the facial nerve are called the "muscles of facial expression" in almost every current textbook of anatomy. True it is that their varying degrees of contraction produce those small alterations of facial contour to which by experience we are highly sensitive, but the muscles are not there primarily to produce facial expressions. They are arranged as circular sphincters and radial dilators around the facial orifices (eye, ear, nose, mouth). In man the sphincter and radial dilators are best developed around the mouth. Contraction of the orbicularis oris compresses the lips into the smallest possible circle. The orbicularis is surrounded by muscles that radiate away like the spokes of a wheel; these muscles are the opponents of the orbicularis. When they all contract simultaneously they stretch open the lips into the widest possible circle. A fact of especial importance to the orthodontist is that the facial musculature is applied over a convex skeleton of bone. Contraction of the muscles causes pressure upon the underlying bones and teeth. The lips press backwards upon incisors and

canines, the modiolus and the buccinator press inwards upon premolars and molars. These pressures are counteracted by that of the floor of the mouth and the tongue within the teeth, and during mastication the bolus is moved and returned between the occlusal surfaces of the teeth by reciprocal contraction of the facial muscles on the one hand and the floor of the mouth and tongue on the other. The buccinator and tongue are important muscles of mastication.

There seems little doubt that inequality of pressure between these opposing forces can result in shift of the teeth in one or other direction. Whether this is the only, or even the chief, cause of malocclusion is not clear. Once malocclusion exists for any reason in the young it will tend to persist or increase by reason of muscle pressures, for a sloping tooth is in unstable equilibrium and separation of the teeth allows greater protrusion of cheek or tongue between them.

That unbalanced muscle actions due to faulty habits can cause malocclusion is almost certain. It is illustrated in a most striking way in certain Abyssinian races, notably the Amhara and Tigrean, whom the writer had opportunity to study closely over a period of four years. Most of these people have gross malocclusion of the type Class II Division I (Angle). They have narrow, highly-arched palates, large central upper incisors that slope forwards with great prominence, and malerupted or impacted wisdom teeth are very common. It is quite rare to find an individual with normal occlusion. So protuberant are the upper incisors that it is only by conscious effort that the lips are held in apposition, and in the relaxed position the upper lip rides up to expose the central incisors, thus giving a false impression of smiling. Equally universal is the habit of chewing with the lips open, the action being accompanied by audible clicking and sucking tongue movements, and *swallowing is usually performed with an open mouth.*

This masticatory habit is universal throughout life, from babyhood to old age. The forward movement of the tongue is unopposed by the lips; conversely the modiolus lies with unwonted pressure against the premolars to prevent the vestibular contents from escaping through the open lips. The incisors are pressed forwards by the tongue, the premolars and molars are pressed inwards by the cheeks. This acquired dental characteristic is not transmitted. The young babies have normal faces with well-arched gums in good contact, but the dental deformity appears in early childhood and progresses into adult life.

These universal chewing habits seem to me to be the undoubted cause of the universal malocclusion, and I cannot help feeling that in Europeans a potent cause of malocclusion may lie in similar faults in chewing rather than in swallowing. Swallowing is a momentary event, chewing a much more prolonged affair, and the forces applied by the tongue and cheeks are much greater in chewing than in swallowing, especially in chewing the harder varieties of food. Perhaps it is fallacious to argue that we swallow more often in twenty-four hours than we chew, because the forces applied to the teeth and alveolar processes are so much greater in chewing, especially if the lips are parted. For with parted lips the forward thrust of the tongue is unopposed.

In the uninhibited Abyssinian, who swallows with teeth and lips widely open, there is, indeed, no forward thrust of the tongue in swallowing, but only in chewing.

In the well-trained European there is a different state of affairs, for if the lips are kept together malocclusion, however caused, encourages an "infantile" type of swallow, for the tongue and lips now meet between the parted teeth. To argue that a persisting infantile type of swallow is the original cause of the malocclusion seems to the writer to be highly speculative and unconvincing, confusing effect with cause.

Summary

(1) Skull movements on the spine are produced chiefly by semispinalis capitis, longus capitis and sternomastoid. The muscles of mastication play no part in these movements.

(2) Opening of the mandible is produced by the digastric and the lateral pterygoid muscles.

(3) The floor of the mouth is shortened by geniohyoid, lengthened by stylohyoid, raised by mylohyoid and lowered by the infrahyoid strap muscles.

(4) Malocclusion is just as likely to be produced by faulty chewing movements as by faulty swallowing movements.

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Section of Urology

President—HUGH DONOVAN, M.B., F.R.C.S., F.R.C.S.Ed.

[*January* 28, 1954—*continued from June "Proceedings"*]

Spontaneous Cure of a Ruptured Bladder

By GEOFFREY PARKER, D.S.O., M.D., F.R.C.S.

THERE are certain injuries and disasters to which men and women are liable which are usually regarded as inevitably fatal, either in a matter of hours, or, more rarely, days, unless surgical intervention be undertaken. Moreover, it is generally taught that operation for their relief and cure is of vital urgency; and yet it seems that this was not always the case.

A diagnosed ruptured ectopic gestation was sometimes treated expectantly and not always with a hopeless prognosis, in the days when simple exploration of the peritoneum carried a very great risk of death from peritonitis.

Cranial fracture also, resulting in the rupture of the middle meningeal artery, may be a self-limiting injury and not necessarily fatal without operation.

Expectant treatment of a perforated gastric or duodenal ulcer, even in the days before continuous gastric aspiration was thought of, did not result in 100% mortality and the successful incision of a perigastric abscess from perforation into the lesser sac or a slow leak, with resulting limited spread of peritonitis, was by no means unknown.

Again, it is seen from time to time in primitive parts of the world that untreated fractures, even with gross displacement of the fragments of the broken ends, may ultimately give a bony union which is functionally, if not anatomically, sound.

Winsbury-White recalls that Morris in 1887 could claim cures of cases of ruptured bladder by the simple use of an indwelling catheter and that the first unsuccessful attempt at suture of a ruptured bladder was made by Willett in 1876.

However, in this day and age, it is axiomatic that a rupture of the bladder, either intra- or extra-peritoneal, calls for immediate surgery, and I am not, of course, claiming that this exceptional case does anything except prove the rule.

Case history.—These are the facts:

A woman of 58 was admitted after a severe motor smash to a hospital, about 60 miles from London, on November 8, 1952. She was suffering from concussion, was noisy and wildly disorientated. An X-ray of the skull showed no fracture, but ribs, 2, 3, 4, 5, and 6 on the left side were fractured, as was also the left wrist.

The story, which must with difficulty be believed, is that she did not develop hæmaturia until one week after admission and this was thought to be due to bruising of the left kidney.

Her general condition in the meanwhile, that is, in the week since admission, had improved, and although she remained disorientated, it was decided to move her to the Woolwich Memorial Hospital where she was much nearer to her home.

On admission, she was found to be running a temperature up to 102° F. and the urine was both heavily blood-stained and infected.

At the request of a colleague, I saw her for the first time on December 15, 1952, four days after admission to Woolwich and thirty-seven days after the accident. There was a tender boggy swelling in the right iliac fossa, which extended up to the lower pole of the right kidney, but faded out in this region, and the maximum tenderness and swelling was clearly extra-peritoneal in the right iliac fossa.

I made a tentative diagnosis of a small tear in the right renal pelvis, or parenchyma, with the resulting slow extravasation of blood and urine down behind the cæcum.

On cystoscopy on the same day, however, I found after repeated washing out of the bladder, not a linear tear, but a large hole in the bladder wall just above the right ureteric orifice and about the size of half a crown (Fig. 1).

I could get the beak of the cystoscope into the hole, but could make out little except bleeding granulation tissue leading into a large unexplorable cavity.

As the general condition of the patient was undoubtedly improving, even in the four days under observation at Woolwich, it seemed to me that immediate surgery was not obviously called for, notwithstanding the certainty of the diagnosis and so I left in a Foley catheter and awaited events.

The mass in the right iliac fossa at once started to get smaller and the temperature dropped, and so, five days later, I did a cystogram, using about 5 oz. of Pyelectan (Fig. 2).

An intravenous pyelogram done some days later showed normal kidney function on both sides.

Nine days later, I cystoscoped her again and found only a large granulating area on the right bladder wall where previously there had been a gaping cavity. I took out the catheter, urine passed normally and she left hospital a week later.

All this happened a year ago. I have seen her from time to time since then, but cystoscoped her to complete my record of this short case ten days ago, the first time since the accident.

There is a small pale radiating scar at the site of this huge tear; so small, in fact, that I think it could

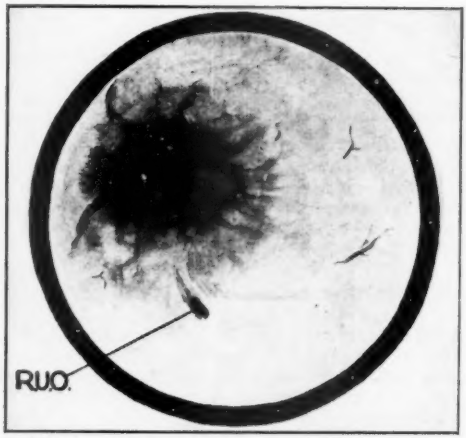

FIG. 1.—Cystoscopic appearance of the rupture.

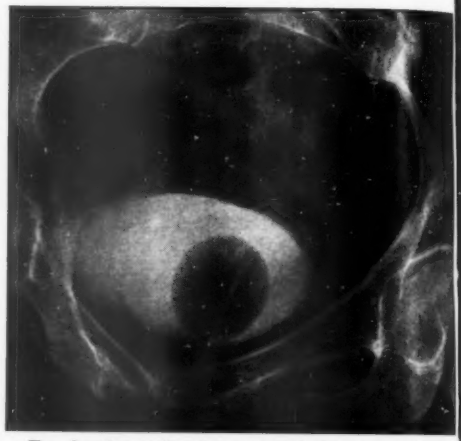

FIG. 2.—X-ray showing extravesical extravasation.

easily be missed, but for the prescience of where to look for it. The patient is free of urinary symptoms, though she has, unfortunately, developed a most severe post-concussional neurasthenia.

Finally, I would repeat that I would not for a moment suggest expectant treatment for the condition of ruptured bladder, but this case, I submit, does show that Nature from time to time has a trick or two up her sleeve, worth at least as much as may be found in any textbook.

Massive Calculi: One Case

Another unusual case is that of a man, aged 66, who was admitted to hospital while I was operating, some weeks ago. I saw him between cases and found him to be suffering from severe painless

FIG. 3.—Untreated calculosis.

hæmaturia. He was comatose but could be roused with difficulty and stony masses were easily palpable in both flanks. A blood urea reported later was 450 mg./100 ml.

The man died in coma an hour later and Fig. 3 is an X-ray taken shortly before his death.

According to his wife, although he had had mild trouble with his water for some years, he had never really had any pain worth mentioning.

One wonders what suffering might have come his way, had he fallen into surgical hands many years earlier.

Mr. Leslie N. Pyrah: It is sometimes possible to deal with instrumental rupture of the bladder by conservative measures. Prior to 1946 I had two instrumental ruptures or tears of the bladder when doing punch prostatectomy. In both these cases I opened the bladder and repaired the rent and the patients recovered. When visiting American clinics in 1946 I was present in the operating theatre when a surgeon ruptured the bladder during a resection of the prostate with the punch. The patient experienced great pain and was given morphia. The surgeon completed the resection, tied in a Foley catheter to which a suction apparatus was attached; the patient was given sulphonamides and penicillin. He made a normal recovery. I have since then myself successfully treated one such case of instrumental rupture conservatively with no untoward after-results. It is necessary to see the patient frequently in the first twenty-four hours until the tear in the bladder is sealed off by blood clot. Persistent local rigidity would indicate the need for cystotomy.

Mr. George Qvist said that he thought this case was more in the nature of a delayed rupture and that the initial injury had caused a severe hæmatoma in the wall of the bladder which had ruptured a few days later. He had seen a case of this type in a patient following myomectomy where damage to the bladder was suspected. A catheter was left in for a week but within three or four hours of removing the catheter the patient died suddenly and autopsy demonstrated a large transverse rupture of the base of the bladder at the site of a hæmatoma. He thought it was important to recognize the occurrence of hæmatoma with delayed rupture both from the clinical point of view and also from the medico-legal aspect and he recalled the recent case in which a patient had had such a lesion in the cæcum and in which an eminent member of their profession had influenced the judge very unfavourably by making an unjustified statement that there *must* have been rigidity present because there was an injury of the cæcum. The whole point, of course, was that there was no rigidity at the commencement and hence the difficulties of the case. It was important to recognize that there might be a considerable hæmatoma in the wall of a hollow viscus which might resolve but which might subsequently rupture and it was often impossible to recognize such a condition clinically in the first few days.

Mr. David Band said when transurethral resection was performed on the neurogenic bladder, there was a very real danger of extravasation. This was especially so when apparently only a minimal resection was required and patients had become debilitated from a prolonged illness. Early recognition of the extravasation and drainage of the space of Retzius was essential. He had also met with the complication of rupture of the tuberculous bladder during cystoscopy under general anæsthesia. In this patient one kidney had been removed for tuberculous disease some years before but there had been deterioration in her condition with frequency and dysuria and finally severe hæmorrhage from the bladder. Several pints of blood were transfused. The bladder capacity had become reduced to 50 ml. and the perforation occurred through the base of a tiny tuberculous ulcer situated at the junction of the roof and floor. The complication was recognized and dealt with by repair of the perforation and intubation of the ureter to the exterior by means of polythene tubing, and so by-passing the bladder which was drained by an indwelling catheter *per urethram*. Some weeks later the remaining ureter was transplanted to the colon by an extra-peritoneal approach.

Mr. A. Jacobs said that though Mr. Parker's second case had survived to a good age in spite of his massive bilateral renal calculi, it should not be inferred that this nullified the possibility of benefit, following operative intervention for this type of calculosis. He was convinced that operation could not only effect marked relief from the severe symptoms which some of the patients experienced especially as a result of infection: but that inexorable renal destruction could sometimes be arrested. Temporary nephrostomy drainage after removing the stones in turn from each kidney was a safety factor and permitted post-operative irrigations with antiseptics or, in the event of debris being left in the kidney, with dissolution agents such as solution G. He thought too gloomy a view was entertained regarding the chances of aiding calculus disease of this nature.

Mr. E. W. Riches said that it was always remarkable to him that the bladder was not perforated more often by diathermy applied to a bladder growth, particularly now that so many were against the radical treatment of carcinoma of the bladder. It happened to him once when he was carrying out deep diathermy in a patient who was old and rather debilitated. He put in a suprapubic tube and drained the extra-vesical space, which was an important part of the procedure. The patient made a good recovery and was alive and well three years later. With regard to the second case he was on the side of the author rather than of Mr. Jacobs. He remembered a man who had such enormous stones in both kidneys that one could feel crepitus through the abdominal wall. He lived for several years until eventually he died from uræmia. If anyone had tried to remove the stones in the stage to which they had developed when first seen they would have formed again very quickly, and his life was probably prolonged by abstention from surgery rather than by its use.

The President recalled the death of a patient from anuria following perforation of the bladder during resection, a number of years ago. He attributed the occurrence to the absorption of water or of a non-isotonic fluid. He felt that a specific answer should be given to the question—should one drain a perforated bladder by catheter?

Mr. Parker, in reply, said that in the case of rupture of the bladder in the woman in Case I, the bladder was ruptured undoubtedly by an impulsive force. Fortunately for her at the time the bladder was practically empty. The indwelling catheter should not be used in a case of ruptured bladder. The treatment, as soon as the patient recovered from the initial shock, should be by surgery. In Case II the man was 66 years old and, according to his wife, he had lived a quiet life and had never had any real pain at all.

[*February* 25, 1954]

DISCUSSION ON ELECTROLYTIC IMBALANCE IN UROLOGY

Dr. D. A. K. Black, Department of Medicine, Manchester Royal Infirmary:

Disorders of the kidney and urinary tract can lead to electrolyte abnormalities by a number of different mechanisms. The electrolyte disturbance may occur somewhat incidentally in a general renal failure, and so be of comparatively minor clinical importance. On the other hand, the electrolyte disturbance may appear so early as to account for the presenting symptoms, and may then continue to dominate the clinical picture; for example, dehydration, acidosis, gross potassium depletion, or demineralization of bone can all arise as complications of urologic disorder. I shall attempt to outline the morbid physiology of the electrolyte disturbances most commonly associated with urological disorders.

In dealing with electrolytes, the kidneys are not concerned to eliminate as much as possible of what is brought to them by the blood, as is done with urea and creatinine; they have the rather more precise task of regulating the excretion of electrolytes so that the body as a whole remains in electrolyte balance. Impairment of this "homœostatic" function of the kidney, as Cannon termed it, may lead sometimes to excessive conservation of different electrolytes, at other times to excessive loss; this type of renal dysfunction may co-exist with quite normal excretory function of the kidneys, as judged by tests of urea excretion. Clinically, homœostatic renal failure expresses itself not as uræmia, but rather as depletion of base (either sodium or potassium, or both), as an acid-base disturbance (usually acidosis), or as an osteomalacia. Of course, "excretory" and "homœostatic" renal failure may co-exist, or one may lead to the other, but there are practical advantages in assessing them, and if possible treating them separately.

(1) *Renal excretory failure.*—This is made manifest by a raised blood urea, or depression of urea clearance, and when severe it gives the clinical syndrome of uræmia. Electrolyte balance may be well maintained, at least until vomiting and diarrhœa occur. Severe electrolyte depletion in patients with no obvious fluid loss, and with only a moderate rise in blood urea, is more likely to be primary than secondary to the uræmia; and appropriate treatment to correct the electrolyte depletion may then improve the renal excretory function, probably by improving the circulation to the kidneys.

(2) *Base depletion.*—Normally the renal tubules reabsorb about 99% of the sodium filtered in the glomeruli, and about 85% of the potassium. This normal conservation of base may be impaired in a number of situations, e.g. during recovery from acute tubular necrosis with anuria, after the relief of urinary obstruction, and in the course of pyelonephritis. Sodium loss or potassium loss may predominate clinically, or they may both be present. Sodium loss, if severe enough, gives the clinical picture of dehydration, with collapsed veins, inelastic skin, and a dry tongue. Potassium loss may lead to muscular weakness or even paralysis and, when severe, to paralytic ileus, and cardiac or respiratory failure.

(3) *Acidosis.*—On a normal diet, the amount of fixed acid produced in the course of metabolism exceeds the amount of fixed base; to maintain a pH of around 7·4 in the extracellular fluids of the body, the kidneys must excrete an excess of anions over cations. The tubules do this by exchanging hydrogen ions for sodium ions, which lowers the pH of the urine directly; and also by forming ammonia, which allows some fixed anion to be excreted in the urine without accompanying fixed base such as sodium or potassium. Damage to the renal tubules can lead to failure of the normal acidification of the urine, so that acid radicles either accumulate in the body, or are excreted in company with fixed base, leading to base depletion. If the urine is exposed to colonic mucosa for a time, reabsorption of chloride in excess of sodium and potassium may undo the previous renal conservation of fixed base; but direct tubular damage by pyelonephritis is probably also concerned in the acidosis which can follow uretero-sigmoidostomy. Occasionally, renal tubular acidosis may be severe enough to be manifest as hyperpnœa; more often, it becomes apparent either through estimation of the alkali reserve, or by contributing to osteomalacia. This type of renal acidosis is distinct from that which arises in renal excretory failure as a result of retention of phosphate and sulphate.

(4) *Osteomalacia.*—The essential lesion in osteomalacia is a failure to calcify bone matrix, the bone matrix being in itself normal, and laid down in the usual way by osteoblasts; the final demonstration of osteomalacia may rest on bone biopsy, which reveals broad "osteoid seams", in which calcium salts have failed to be deposited. Failure to calcify bone matrix indicates a deficiency in the local tissue fluid of phosphorus, or calcium, or both; or it may arise in acidosis, which seems to prevent normal mineralization of bone at levels of calcium and phosphorus which would normally not be associated with osteomalacia. When the renal tubules are damaged, they may excrete

excessive amounts of phosphate, leading to a low plasma phosphate, and so to "low-phosphate osteomalacia"; but the osteomalacia which can complicate urological disorders is more commonly related to acidosis. Clinically, osteomalacia may present as diffuse bone-pains, incomplete pathological fractures, or increased curvature of the spine with loss of height.

When excretory renal failure is present, bone symptoms may arise by an entirely different mechanism, in which the sequence is something as follows—phosphate retention gives a low serum calcium, probably by inhibiting calcium absorption from the gut; the low serum calcium stimulates parathyroid hyperplasia, which promotes increased osteoclastic activity; this removes the whole structure of bone, both matrix and deposited salts, so that we have renal osteitis fibrosa, with a general poverty of bone structure, but no broad seams of uncalcified matrix. The distinction between osteomalacia and renal osteitis fibrosa is far from academic, as intensive alkali treatment is indicated in osteomalacia, and has given good results; whereas renal osteitis fibrosa is probably not responsive to treatment.

Thus there is a variety of metabolic distortions which may be occasioned by renal dysfunction. This could perhaps be regarded as a sort of back-handed compliment to the versatility of renal performance in health; but in its practical bearing it compels us to the somewhat trite conclusion that in this field, as in so many others, each patient is a separate problem, demanding individual management.

Mr. L. N. Pyrah, Department of Urology, University of Leeds, Leeds General Infirmary and St. James's Hospital, Leeds:

My brief in this discussion is to indicate those surgical urological conditions in which electrolyte studies are of very great importance in the treatment of the patient. Table I shows a list of surgical conditions in which we have found imbalance to occur.

TABLE I.—UROLOGICAL CONDITIONS IN WHICH ELECTROLYTE IMBALANCE MAY OCCUR

1. *Anuria*
2. *Kidneys in which tubular damage predominates*
   - (A) After uretero-colic anastomosis.
   - (B) Renal acidosis.
3. *Kidneys badly damaged by disease*
   - (A) Calculous disease.
   - (B) Bilateral infected kidneys { Tuberculous / Pyogenic
   - (C) Bilateral hydronephrosis. Hydronephrosis of solitary surviving kidney.
   - (D) Congenital cystic kidney.
4. *Obstruction in the lower urinary tract*
   - (A) Chronic retention with overflow.
   - (B) Sudden release of obstruction.

ANURIA

I will refer briefly to only one type of anuria, namely the renal type. Fig. 1 shows the clinical course of a case of anuria following abortion. The anuric phase lasted a fortnight. I would particularly draw attention to the early diuretic phase, for it is during this phase that the persisting tubular damage may allow a dangerous loss of water and electrolytes, the most important being chloride, sodium and potassium. In this case there was a loss of chloride and of sodium; they were replaced as sodium chloride by supplementation. There was also a high output of water, but not as high as in many cases. Exactly similar high losses of water and electrolytes may occur in patients with badly damaged kidneys when obstruction is suddenly relieved; such obstruction may be due to an enlarged prostate causing retention, to a bilateral hydronephrosis, or to an impacted ureteric calculus (the other kidney also being damaged or absent).

KIDNEYS IN WHICH TUBULAR DAMAGE PREDOMINATES

These cases fall into two groups. Firstly the *idiopathic group* which we call *renal* acidosis; secondly the group of renal abnormalities which may follow uretero-colic anastomosis.

Renal Acidosis

There is a group of conditions in which renal tubular damage of unknown cause is the primary pathology. These conditions present with different characteristics in different age groups. Because the tubules are damaged, the kidney cannot conserve base and calcium will eventually be mobilized from the bones, leading to a type of rickets in adolescents, or to a varying degree of osteomalacia in adults, also to a deposition of calcium in the tubules at any age—a condition which we call nephrocalcinosis. The loss of base in these patients leads to a hyperchloræmic acidosis. The essential part of the treatment is to supply base. I will refer to one case of each group—infants, adolescents and adults.

(1) *Idiopathic renal acidosis of infants.*—There is a group of cases of renal acidosis in infants under 1 year, which usually present with a history of failure to gain weight, dehydration, hypotonia and polyuria. Investigation reveals findings which are typically shown in the case in Table II.

TABLE II.—IDIOPATHIC RENAL ACIDOSIS IN AN INFANT OF 8 MONTHS (P. S., FEMALE)
The Condition Responded to Treatment by Alkalis. Blood Chemistry Before Treatment and at End of Treatment

Normal values			14.8.53	12.2.54
560–620	Chloride	mg.NaCl%	610	580
96–106		mEq/l	104	99
55–70	CO_2 c.p.	Vol.%	37	53
24·8–31·5		mEq/l	16·7	24
305–325	Sodium	mg. Vol.%	300	300
132–141		mEq/l	130·5	130·5
16–20	Potassium	mg. Vol.%	20	16·8
4·1–5·1		mEq/l	5·1	4·3
10–20	Urea	mg.N₂%	11·8	21
2–4	Phosphorus	mg.%	4·5	4·3
1·16–2·32		mEq/l	2·6	2·5
9–11	Calcium	mg.%	9·8	9·8
		mEq/l	4·9	4·9
	Urine pH		7·7	6·5

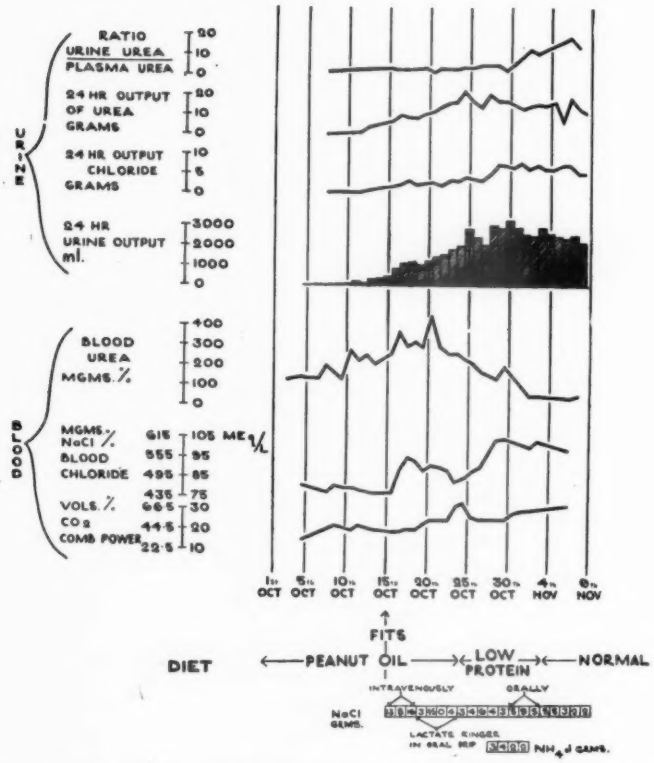

FIG. 1.—Mrs. E. G., aged 31. Blood chemistry and urine chemistry and urinary output of a case of anuria following septic abortion.

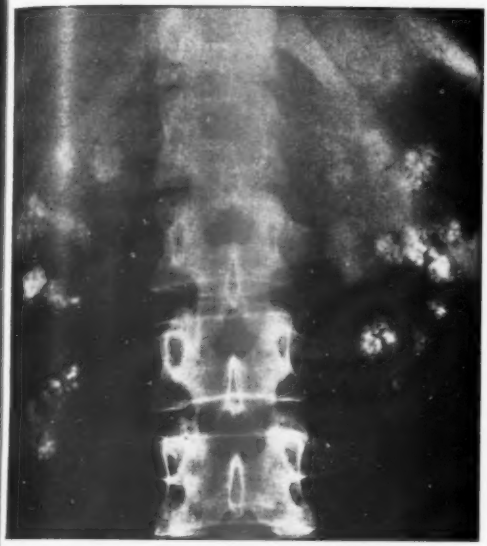

FIG. 2.—Intravenous pyelograms of kidneys showing nephrocalcinosis of both kidneys secondary to renal acidosis.

There was a high normal chloride and a low CO_2 combining power; the urinary pH was 7·7, the urine containing an excess of sodium bicarbonate. The final column shows the blood chemistry corrected after six months' treatment with base. After that time the child had put on weight and the symptoms had disappeared. The child was treated with a daily dose of potassium citrate 2 grammes, and sodium citrate 4 grammes in 4 oz. of water, given in doses of 1 oz. four times a day. The dose will vary from case to case according to the effect produced.

(2) *Cases in adolescence.*—Albright and his co-workers (1940) reported a case of renal acidosis in which the loss of calcium through the kidney had led to nephrocalcinosis and also to a type of rickets. This was treated firstly by giving base, and secondly by giving calcium, phosphorus and vitamin D. There was dramatic improvement. The base was given in the form of Shohl's solution (citric acid —140 grammes, sodium citrate—98 grammes, water—1 litre: 50 to 100 ml. daily, according to therapeutic response). I myself have not seen a case in this group.

(3) *Renal acidosis in adults.*—

Fig. 2 shows the radiograph of the kidneys in one such case, a woman aged 43, who presented with recurrent attacks of renal colic with passage of small stones, for five years; the urine was sterile. The radiograph illustrates the typical distribution of the pyramidal calcification in nephrocalcinosis. At a later stage her main complaints were an unquenchable thirst, a nasty taste in the mouth and weakness of both arms and legs, so that she was unable to do her work. The blood chemistry, shown in Table III, revealed a hyperchloræmic acidosis (an elevation of blood chlorides and a loss of base shown by a reduction of CO_2 combining power) and also a low potassium; the low potassium was responsible for the weakness of the arms and legs.

She was treated by reducing the chloride in the diet, and by Shohl's solution, but because the potassium was low the sodium citrate was partly replaced by the potassium citrate. The potassium was not brought completely to normal but she felt better with it at that level rather than at a higher value. She is now free from symptoms and able to work, but the nephrocalcinosis has not yet altered.

TABLE III.—CASE OF NEPHROCALCINOSIS DUE TO RENAL ACIDOSIS, R. H. (FEMALE, AGED 43)
Table shows blood chemistry at various dates. She was treated by modified Shohl's solution, potassium citrate partly replacing the sodium citrate

Normal values		18.4.53	13.5.53	16.5.53	19.5.53	2.9.53
40–47	Hæmatocrit	41·5	41	42·5	40	39·5
6·5–7·5	Plasma protein gm.%	8·85	8·53	8·6	8·45	8·3
10–20	Urea mg. N_2 %	13	12	27	35	13
96–106	Chloride mEq/l	115	109	104	108	105
24·8–31·5	CO_2 c.p. mEq/l	16·9	26·2	25·9	25·5	22·5
132–141	Sodium mEq/l	139	145·5	137	139	140
4·1–5·1	Potassium mEq/l	2·8	3·5	4·19	3·7	3·88
95–102	Potassium R.B.C. mEq/l	92	100	97·7	96·5	97·5
1·16–2·32	Phosphorus mEq/l	3·35	3·48	4·38	3·23	4·3
9–11	Calcium	10·4	10·9	—	—	10·1

Uretero-colic Anastomosis

A good deal is now known about the imbalance which occurs following uretero-colic anastomosis and as this subject has also been discussed recently I shall only briefly refer to it. (Parsons, Powell and Pyrah, 1952; Parsons, Pyrah, Powell, Reed and Spiers, 1952; Pyrah, 1954.)

We know that chlorine ions and sodium ions are absorbed from colonic urine across the colonic mucosa, chlorine ions being absorbed to a greater extent than sodium ions. If the renal tubules have been damaged by infection, a hyperchloræmic acidosis will result, and this has been found to be present in a varying but often high percentage of cases having this operation. This imbalance can usually be controlled by excluding sodium chloride taken as a condiment in the diet, and by taking sodium bicarbonate daily. In cases which do not observe these rules, or if the renal damage increases, they may develop the severer degrees of imbalance characterized by extreme dehydration leading to anuria, severe acidosis leading to coma, and dangerous degrees of hypokaliæmia.

Kidneys Extensively Damaged by Disease

I will now refer to the group in which the kidneys were extensively damaged by disease. We have seen many examples of this resulting from stone and hydronephrosis, but I will refer only to two cases of extensive calculus disease. With very severe renal disease, both glomerular and tubular damage occur, and there may be retention of some electrolytes and excessive loss of others, thus leading to chemical imbalance. This imbalance must be recognized and corrected; we have found that the correction itself may lead to improved renal function.

Case I.—M. G., female, aged 72. Bilateral renal calculi. The patient complained of backache for ten years. Three years ago she had passed some stones and six months ago she had had a profuse hæmaturia. And then came the decline. In the few months before admission there had been a marked deterioration of general health with loss of appetite, headaches, thirst, loss of weight and slight mental changes which were attributed to old age. Fig. 3 shows the very large staghorn calculi. The urine was heavily infected. On physical examination there was severe dehydration as evidenced by a dry tongue and loss of skin turgor; she had acidotic breathing and slight mental confusion. This was the picture of a case of bad renal function in which one would expect to find electrolyte imbalance.

Fig. 3.—M. G., female, aged 72. Huge, bilateral, staghorn calculi, gross renal dysfunction, electrolyte imbalance.

The figures (Table IV) show a low plasma chloride, which in the short space of three days lost a further 5 mEq/l; a low CO_2 combining power, and a low sodium, which also fell further in three days. The potassium was normal but falling. The urea nitrogen, though high, gave no indication of the extent of the chemical imbalance.

The patient declined treatment, and died at home ten days later, from what was described as a terminal coronary thrombosis, but which may have been a potassium deficiency. This was, in fact, the final stage of a patient whose kidneys were severely damaged by stone and infection.

TABLE IV.—BLOOD CHEMISTRY OF CASE I, M. G. (FEMALE, AGED 72). THERE IS A SODIUM CHLORIDE DEFICIT AND AN ACIDOSIS

Normal values			25.12.53	28.12.53
560–620	Chloride	mg. NaCl%	500	470
96–106		mEq/l	85·5	80·5
55–70	CO_2 c.p.	Vol.%	48	47
24·8–31·5		mEq/l	21·7	21·3
305–325	Sodium	mg. Vol.%	280	275
132–141		mEq/l	122	119
16–20	Potassium	mg. Vol.%	18	15·5
4·1–5·1		mEq/l	4·6	4·0
10–20	Urea	mg. N_2%	56·1	
2–4	Phosphorus	mg.%	4·2	4·2
1·16–2·32		mEq/l	2·44	2·44
9–11	Calcium	mg.	10·3	9·9
		mEq/l	5·15	4·95

Case II.—D. L. R., male, aged 56. This case illustrates the management of a patient, who had only one kidney which was the seat of a large calculous pyonephrosis, and who had uræmia.

The patient had had his right kidney removed for calculous pyonephrosis in Vienna in 1929. Since then, he had had occasional discomfort in the left kidney, and hæmaturia. He was first seen by me in July 1947 by which time he was known to have had for four or five years stones in the left kidney with intermittent urinary infection (Fig. 4). The stones were in the pelvis and lower calyces and there was a moderate grade of hydronephrosis. The blood urea nitrogen was 35·6 mg.%. The patient was unwilling to consider any operative interference. I followed this patient between 1947 and 1951 and the stones slowly increased in size, the hydronephrotic changes were more marked and the blood urea nitrogen was slowly rising. He was still able to work and was almost symptom-free. And then came the change.

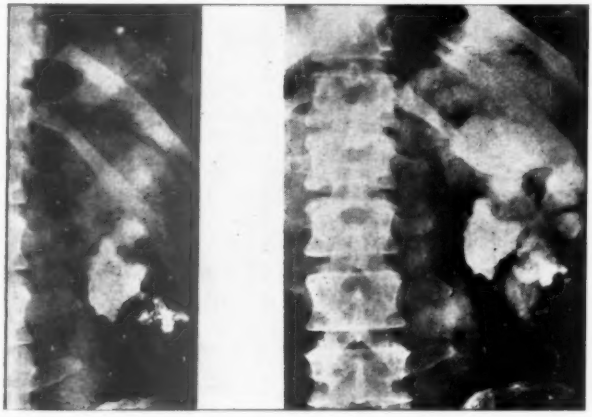

FIG. 4.—D. L. R., male, aged 56. Massive calculi in solitary kidney with infected hydronephrosis; on the left, control X-ray (30.6.47) showing multiple calculi; on the right, intravenous pyelogram (30.6.47) 25 mins. picture showing hydronephrosis.

During the next twelve months he gradually lost energy and found mental concentration increasingly difficult. His appetite began to fail. And then came the crisis! In June 1952 he spent a holiday abroad in a heat-wave; he lost his appetite, developed a nasty taste in the mouth and also severe thirst, and felt generally ill. On medical advice, he was told to reduce his intake of fluids and he became gradually worse. It was only with difficulty that he made the journey back to Leeds where he was admitted to hospital.

When I saw him the man was critically ill; he had a dry mouth, he could only speak slowly and with difficulty, cerebration was slow and dulled, he had no pain, the skin was dry and there was an absence of normal skin turgor; he had severe acidotic (Küssmaul's) breathing. The temperature was 102° F. and he had had some rigors; the pulse-rate was 120. He was still passing urine which was turbid with a heavy content of pus. There again was the clinical picture of gross renal impairment, urinary infection and electrolyte imbalance as shown by *dehydration, acidotic breathing* and *mental impairment*. His condition was so poor that it did not seem that he could recover.

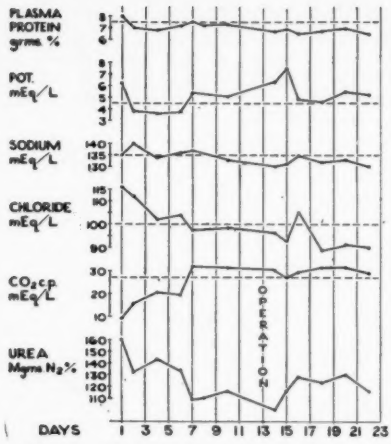

FIG. 5.—D. L. R., male, aged 56. Pre-operative and post-operative blood chemistry. On admission he had a hyperchloræmic acidosis, a grossly elevated blood urea nitrogen and dehydration.

Fig. 5 shows the blood chemistry on admission to hospital and subsequently (23.7.52). He had a hyperchloræmic acidosis with elevation of the potassium and plasma phosphorus; the blood urea nitrogen was 160 mg.% The high plasma protein suggested that there was, in addition to a water depletion, probably some sodium depletion although the blood sodium level appeared to be within normal limits.

What was to be the treatment? Any immediate operation was out of the question. If he was to survive, treatment had to be directed to the following five defects which collectively were responsible for his plight:

(1) *The immediate correction of the dehydration and acidosis.*—He was allowed water by mouth, and he was also given an intravenous infusion of 1½ litres one-sixth molar sodium lactate. This treatment should produce a beneficial effect on the dehydration, secondly on the sodium depletion and thirdly on the acidosis, and Fig. 5 illustrates this. Clinically, thirst was reduced, his cerebration improved, and his acidotic breathing disappeared.

(2) *The nitrogen retention.*—With improvement in the urinary output consequent on the relief of the dehydration, the blood urea nitrogen commenced to fall. In order to reduce still further the blood urea level, a high-glucose, low-protein diet was given; 400 grammes of glucose were given daily and the only protein administered daily was that contained in one pint of milk. Although on admission he was in a state of malnutrition, this diet was sufficient for stabilization.

(3) *Electrolyte loss.*—Consequent upon the relief of dehydration and the resulting output by the badly damaged kidney, there is a tendency for the loss of all electrolytes. For evidence that this is occurring one has to watch the electrolyte levels of the blood plasma, and the electrolyte concentrations in the urine. Supplementation which is then required is estimated on these findings.

(4) *Infection.*—This was treated by aureomycin. Sulphonamides and streptomycin are contra-indicated in cases of defective renal function.

(5) *Anæmia.*—The hæmatocrit was low. The correction of anæmia which often accompanies severe renal damage, I believe should be performed cautiously, but it is necessary to fit the patient for operation. Packed cells are to be preferred to whole blood. This patient received two pints of packed cells.

We had now reached the 10th, 11th and 12th days; the patient's condition would improve no further. The clinical diagnosis was a calculous pyonephrosis. What was to be done? The only hope of improving renal function was to remove the stone and drain the kidney. It was realized that the risk was high.

On the twelfth day after admission the left kidney was explored and found to be the seat of a tense pyonephrosis. The stones were removed across the renal pelvis and the remainder of the stones, by opening into the distended lowest calyx. The kidney, when collapsed, was a series of sacs, the walls of which were no thicker than 3 mm., except near the upper pole where there was a small area of thicker renal tissue. The amount of renal tissue in this man's kidney was the *least* that I have ever met with in a patient who survived.

In the post-operative period of such a gravely ill patient, operation on the already severely damaged kidney may produce an even further deterioration in renal function, but consequent upon the relief of obstruction this should be temporary in nature. Attention should be paid to the following principles:

(1) Protein catabolism should be suppressed to the minimum, and because of possible gastro-intestinal upset consequent upon anæsthesia we have used 20% or 40% glucose administered by a polythene tube, inserted through the basilic vein and passing upwards into the great veins in the upper part of the thorax, where immediate dilution of the irritant glucose takes place. By the use of 2½ litres of 20% glucose, protein catabolism is suppressed and also adequate hydration obtained.

(2) A careful check should be made on the plasma-electrolyte concentration so that any retention of electrolytes, notably potassium, can be immediately dealt with. In this patient, on the second post-operative day, the serum potassium had risen to a dangerously high point for an oliguric patient, necessitating the use of an ion-exchange resin. He was given 80 grammes Zeo-Carb. 225/Na phase, by mouth. The amount of this substance to be used is estimated by the initial level of potassium and by the fall which is produced following its administration. In this patient there was a dramatic fall to normal levels.

(3) At a later stage post-operatively when the oliguria has gone, it is essential that the urinary electrolyte levels should be estimated to guard against excessive loss, and if this occurs to supply the appropriate supplementation.

The patient made slow progress. On the sixth day he was given a low-protein diet. From this point he progressed well and one month after operation he was walking about in the ward, eating a modified diet and

gaining weight. The kidney wound healed. He was advised to take a low-protein and low-salt diet and 4 grammes of sodium bicarbonate daily. Three months after operation he returned to full-time work and is known to be working now.

Obstruction in the Lower Urinary Tract

Prostatic obstruction.—In cases of *chronic retention of urine with overflow* cases of electrolyte imbalance occur and occasionally also in cases admitted with acute retention where early operation has been performed. Some of these are admitted with severe azotæmia, but the blood urea level alone should never be allowed to give a feeling of security, since marked alterations in the internal environment may be present with little or no urea retention. We have found three groups of cases, though there is much overlapping; these are (1) the diuretic group, (2) a group having water and electrolyte retention, and (3) a mixed group commonly associated with acidosis which we have called a uræmic group (Parsons, 1954).

An example of the diuretic group.—The case was a man (R. E.) aged 54, who was first admitted to the Medical Department on account of loss of appetite, nausea, vomiting and epigastric pain and was then found to have chronic retention of urine with overflow, whereupon he was transferred to the Urological Department. The blood chemistry before catheterization showed a high blood urea nitrogen (140 mg.%), the chloride and the sodium were normal, the CO_2 combining power was low (17·5 mEq/l), and the blood potassium was raised.

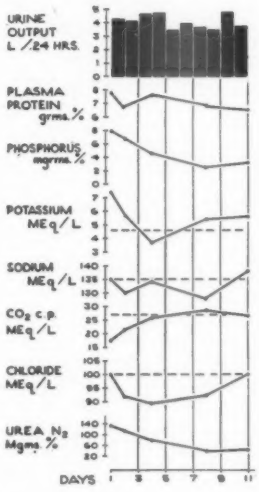

FIG. 6.—R. E., male, aged 54. Simple enlargement of the prostate. Chronic retention of the urine with overflow. Electrolyte imbalance (diuretic group).

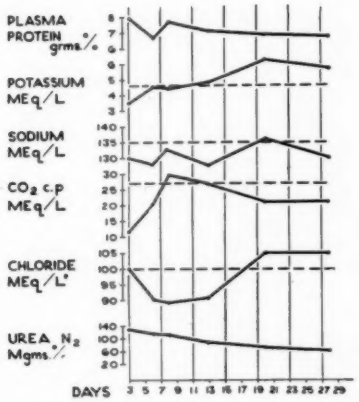

FIG. 7.—A. K., male, aged 57. Case of prostate obstruction with overflow retention. Severe electrolyte imbalance (uræmic group).

After catheterization the urinary output was high, lasting for ten days; but no dehydration resulted, as fluid intake was voluntarily increased to balance output; but together with this high water output he also lost electrolytes, and Fig. 6 shows the fall in the blood levels of these. Daily examination of the urine showed that the loss of electrolytes continued, and supplementation was therefore necessary. By the eleventh day he was balanced. Retropubic prostatectomy was performed on the sixteenth day without complications.

Uræmic group.—The cases of severest imbalance have occurred in this group. The following case is an example.

Male (A. K.), aged 57. On admission he was confused and unable to give complete history for several days. He had had urinary incontinence for six months and in the weeks preceding admission he had found difficulty in concentration and difficulty in carrying on his business. On admission he was found to have a full bladder, due to chronic retention. A catheter was passed and for the first seventy-two hours urinary output exceeded intake and his general condition became slowly worse. Although output exceeded intake for the first two or three days, this was not maintained as in the diuretic group. The blood chemistry on Day 3 showed a much raised blood urea nitrogen, the chloride was normal, the CO_2 combining power was very low, and the sodium and potassium slightly low. *On account of his high blood urea* he was treated by intra-gastric

drip with an anuric diet together with electrolyte supplementation. Fig. 7 shows the gradual improvement. He was not fit for operation until the twenty-eighth day when he had a perurethral prostatectomy and made a good recovery.

SUMMARY

The principal causes of electrolyte imbalance in surgical urological conditions have been outlined and some examples have been given.

Of special importance to the surgeon are those cases in which the kidneys have been badly damaged by disease (calculus disease, hydronephrosis, infection, chronic prostatic obstruction). In such cases electrolyte imbalance may be expected to occur from the known clinical history of the patient. Imbalance may present for the first time as the syndrome of dehydration, acidotic breathing and mental impairment. Examples have been given of some of the conditions producing imbalance, and the management of such cases has been outlined. When operation has to be performed, in general it should not be simply a life-saving procedure such as nephrostomy or cystotomy. Such an operation could never be final and would admit further infection. The operation in general should be planned to relieve the obstruction (kidney, ureter, or prostate) after the internal environment has been stabilized by biochemical means.

Acknowledgments.—I desire to thank Dr. E. C. Allibone for permission to record the case of renal acidosis in infancy, Dr. F. M. Parsons for the biochemical work in the case of anuria and in the case of D. L. R., and Mr. J. Hainsworth for the photographic work.

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Dr. J. A. Black said that he had studied 9 cases at The Hospital for Sick Children, Great Ormond Street, and one case under Mr. Innes Williams at the Institute of Urology. There were 5 fatal cases of congenital urethral obstruction. Operation was done in each case, in the youngest at the age of 9 days and in the oldest at the age of 3 years. Deaths occurred from two to fourteen or fifteen days after operation, and was preceded by gradual deterioration or sudden collapse. In all cases the blood urea fell after the operation, and the serum potassium level fell in a similar manner, often to abnormally low levels. Perineal urethrostomies were done in 4 cases and bilateral pyelostomy in 1 case.

What was the cause of the electrolyte disturbance? Was it due to the mechanical operation of decompression or was it, in fact, the result of the operation itself? In order to try to separate the possible factors some cases with unilateral hydronephrosis were studied, using the normal side as a control. A conservative operation was done in each case and the urine was collected from each kidney separately. The most severely damaged kidney showed an inability to change its electrolyte output and was therefore unable to conserve sodium or potassium during the various phases of the post-operative period. Therefore it was possible that greater post-operative electrolyte losses might occur with the severe bilateral hydronephrosis found in cases of congenital urethral obstruction.

Mr. J. N. Ward-McQuaid summarized the results of an investigation in which 13 patients with paralytic ileus had been studied in an attempt to discover the relation of adrenocortical hyperactivity and of potassium and chloride deficiency to the development of this condition.

There was evidence of adrenocortical hyperactivity during ileus, with abnormal prolongation of the post-operative eosinopenia, excessive potassium loss in the urine, and retention of water, sodium and chloride. On recovery, the eosinophil count rose, and there was a diuresis of water, sodium and chloride and a retention of potassium.

Evidence of potassium deficiency rested on excessive loss during ileus and retention after recovery: it was confirmed by electrocardiograph changes. In 5 out of 10 patients the plasma potassium was low (Streeten and Ward-McQuaid, 1952). There was strong experimental evidence that potassium deficiency produced a state resembling ileus in rats and dogs. There were difficulties in accepting the view that the gastro-intestinal features of ileus in man were due to potassium deficiency following excessive adreno-cortical activity. The work of Dr. D. A. K. Black on experimental potassium depletion in man was well known, and relatively few symptoms followed moderate depletion.

From the practical aspect, however, relatively small doses of intravenous potassium did help patients with ileus. He had used the two Darrow's solutions containing some 40 mEq/litre K, and had also found the weaker gastric and intestinal aspiration replacement solutions (Cooke and Crowley, 1952) of great value. He also wished to know which was considered to be a safe level for the blood urea and for urinary output before intravenous potassium might be given.

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Mr. B. H. Page suggested that one way of grappling with the electrolyte problem was to become familiar with some of the normal figures, and he exhibited a card (Table I) which he had found useful, setting out the normal values of the main electrolytic constituents of the plasma and their concentrations in some of the solutions used in intravenous therapy, expressed in mg./100 ml. and in mEq./litre. This helped one better to appreciate and to correct abnormalities.

TABLE I.—ELECTROLYTES IN I.V. SOLUTIONS, &c.

	A, mg./100 ml. B, mEq/litre									
	Na+		K+		Ca++		Cl−		HCO_3−	
	A	B	A	B	A	B	A	B	Vol.	B
Blood plasma	326	142	19·5	5	10	5	360	103	60	27
Normal saline	354	154	0	0	0	0	540	154	0	0
Hartmann..	295	130	15·6	4	6	3	382	109	62·5	28
1/6 M lactate	385	167	0	0	0	0	0	0	370	167
*Gastric replacement ..	143	63	66·3	17			524·2	150		
*Intestinal replacement ..	113·4	50	46·8	12			394·5	100	110	50

*N.B. Never faster than 500 ml./hr.

Mr. D. Innes Williams said that many urologists would be glad of some guidance as to when all these electrolyte studies should be performed, since few hospitals had adequate biochemical facilities for the full investigation of every case.

One point might be mentioned in connexion with renal acidosis: this was a "medical" disease which normally had nothing to do with the urologist but now and then was presented to him because there were white cells and organisms in the urine. Children suffering from this disease might therefore be referred for investigation of recurrent urinary infection; they were often dehydrated, almost always constipated and rather ill despite a low blood urea. Only routine electrolyte studies would give the correct diagnosis in such cases.

Dr. M. G. Good: If no biochemist is available use could be made of the electrocardiogram, which allows the diagnosis both of hyperpotassæmia and hypopotassæmia (Bickel and Plattner, 1952).

With regard to infections of the genito-urinary tract it is pertinent to point out that the efficacy of antibiotics and bacteriostatics in general depends to a material extent on the pH of the urine, in dependence on the general chemical nature of the drug. It has been shown that a slightly *alkaline* pH *increases* the permeability of microbes for *cationic* inhibitors (e.g. streptomycin, sulpha drugs), and hence its bacteriostatic effect, whereas an *acid* pH *diminishes* permeability and toxicity. Conversely in *anionic* substances (e.g. penicillin) the inhibitory effect is *increased* by an *acid* pH, and *decreased* by an *alkaline* reaction of the vehicle (Good, 1951, 1953, 1954). By adjusting the pH of the urine according to the bacteriostatic, urologists are in the favourable position easily to enhance their therapeutic results in pyelonephritis, cystitis, &c.

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Mr. E. W. Riches said that it was evident that the urologist of the future would have to be a biochemist. It would ease the strain on the memory if there could be uniformity in the expression of chemical values, and he made a plea for the general adoption of milli-equivalents per litre rather than milligrammes per cent; there would then be only one set of figures to remember.

Mr. Russell Johnson said it had been impressed on him that potassium was withdrawn from the plasma when glycogen was synthesized as a result of glucose administration. He wished to ask Dr. D. A. K. Black whether this was a significant factor in case management or merely a theoretical point.

His second question concerned hyperchloræmic acidosis. He had under supervision a man aged about 35 with nephrocalcinosis in whom no biochemical abnormality had been found or history of excessive vitamin administration been elicited. The kidney changes now seemed to be static and he was tempted to suggest that they were the result of a past episode of hyperchloræmic acidosis: could this be a temporary condition?

Mr. Pyrah, in reply, said that in his Department they had not so far had cases of urinary obstruction with associated electrolyte imbalance in small children, but he could imagine the difficulties in obtaining blood. It was his view that in every urological department, particularly in this period of rapid change, biochemical assistance should be available within the department, preferably closely linked to the Department of Chemical Pathology. Many cases which presented difficulty were admitted to hospital at the week-end or during the night and required urgent biochemical attention; preferably the biochemist should also be a clinician. It was important that team-work should be established between clinician and biochemical worker.

Nephrocalcinosis could arise from several different causes in addition to renal acidosis and biochemical methods would, in general, allow the cause of the nephrocalcinosis to be worked out. Amongst the causes of nephrocalcinosis were hyperparathyroidism, vitamin D poisoning, sarcoidosis and excessive ingestion of alkali.

Dr. D. A. K. Black, in reply to the question whether potassium could safely be administered when the blood urea was below a certain level, and the urine volume adequate, said that neither of these criteria would indicate absolutely that potassium could be freely given. Renal tubular insufficiency could be present with a normal blood urea, and urine volume, but with abnormal potassium elimination. Rather than lay down arbitrary figures for blood urea and urine volume, he would stress the necessity for close biochemical control in any patient receiving intravenous potassium. Electrocardiographic control was of value, but one incident has been reported in which a team of workers were giving potassium, and watching the ECG tracing. They suddenly noticed a change in the T-waves, and when they turned to the patient they found that he had collapsed and died.

He was himself perhaps unduly conservative in his attitude to intravenous potassium. Evidence was accumulating that potassium deficiency had got a lot to do with paralytic ileus, and one should be prepared to give potassium by the intravenous route to a patient with ileus, if there was no other route available.

He mentioned that glucose tolerance tests had been known to precipitate attacks of paralysis in patients with potassium depletion; this was probably related to the entry of potassium into cells in association with glycogen formation.

[*March* 25, 1954]

The following Cases and Specimens were shown:

Right Nephrectomies for Hæmaturia — Strange Calculus Constellations (Two Cases).—Mr. A. WILFRID ADAMS.

Ureterograms.—Mr. G. C. TRESIDDER.

Obstructed Hydronephrosis Following Retrograde Pyelogram.—Mr. A. W. BADENOCH.

Multiple Cysts of the Kidney with Stone Formation.—Mr. J. N. WARD-MCQUAID (for Mr. A. W. BADENOCH).

(1) Spontaneous Rupture Hydronephrosis (with Large Papilloma Previously Symptomless in a Woman of 30). (2) Misleading Aortograms: (a) Hypernephroma. (b) Solid Transitional-cell Carcinoma.—Mr. HENRY CLARKE.

X-rays and Drawing of an Unusual Cystogram Due to a Pelvic Tumour.—Mr. R. A. MOGG.

Spontaneous Rupture of Myolipoma of Kidney.—Mr. C. L. O. MACALISTER.

Metastasis in Penis (Corpora Cavernosa) Subsequent to Cystectomy for Bladder Carcinoma.—Mr. ARTHUR JACOBS.

Squamous-cell Carcinoma Arising in a Cyst of the Urachus.—Mr. RICHARD E. SHAW.

Operation for Aberrant Ureter with Unusual Complications.—Mr. T. L. CHAPMAN.

Massive Vesical Calculus—4¼ lb.—Mr. A. CAVENDISH.

Section of Neurology

President—E. A. CARMICHAEL, C.B.E., F.R.C.P.

[*January* 7, 1954]

DISCUSSION ON VOLITIONAL MOVEMENT

Dr. Macdonald Critchley:

> Anaxagoras indeed asserts that it is his possession of hands that makes man the most intelligent of the animals; but surely the reasonable point of view is that it is because he is the most intelligent animal that he has got hands. Hands are an instrument; and Nature, like a sensible human being, always assigns an organ to the animal that can use it. . . . Now the hand would appear to be not one single instrument but many, as it were an instrument that represents many instruments. Thus it is to that animal (*viz.* man) which has the capability for acquiring the greatest number of crafts that Nature has given that instrument (*viz.* the hand) whose range of uses is the most extensive.
>
> ARISTOTLE,
> *De partibus animalium*, IV, X.

Our knowledge of volitional movement has been erected upon a tripod of clinical observation, experimental physiology, and psychology. Our present-day nomenclature needs revision; "voluntary", "volitional", "willed", "purposive", "purposeful", "deliberate", have been inappropriately used as if synonymous, and they have been contrasted with "involuntary", "unwilled", "spontaneous", or "pseudo-spontaneous" activities. Still more unjustified is a careless reference to volitional movements as though they were mediated specifically along pyramidal pathways, initiated by the cells of the motor cortex, and comparable with the effects of electrical stimulation. The pitfalls of loose thinking upon such topics have been disclosed in great detail during the past fifteen years.

Some elements of the problem of volitional movement have at times been forgotten, as, for instance, the fact that there are degrees of volition. We are too apt to assume that the voluntary component of movement is a straightforward mental phenomenon. On the face of it, the attributive "volitional" implies that action has taken place as the result of some operation of the will. This leads us to the question of what is meant by volition and at once we find ourselves in the deep waters of philosophy. Immediately we see the fallacy of making an artificial antithesis between voluntary and involuntary movement, or between voluntary and non-voluntary movement. To choose the term "volitional" helps only a little. The phenomenon of volition remains in all its complexity. William James (1892) believed that the forerunner of a voluntary act lies in a psychic state which comprises an anticipatory image of the sensorial consequences of movements (that is, the motor-cue) together with (on certain occasions) what he called a fiat, in other words, a decision, express consent, or mandate, that these sensorial consequences should be actualized. Then, too, the act of mental consent necessitates the neutralization of antagonistic or contrary or inhibitory ideas. Volitional action may comprise action after deliberation, with all its varying types and methods of decision—attainment. William James went on to say that at this point the pyschology of volition properly stops, and that the movements which follow are exclusively physiological phenomena. In his own words: "Volition is a psychic or moral fact pure and simple and is absolutely completed when the stable state of the idea is there. The supervention of motion is a supernumerary phenomenon depending on executive ganglia whose function lies outside the mind. If the ganglia work duly, the act occurs perfectly. If they work, but work wrongly, we have St. Vitus's dance, locomotor ataxy, motor aphasia, or minor degrees of awkwardness. If they don't work at all, the act fails altogether, and we say the man is paralysed. He may make a tremendous effort, and contract the other muscles of the body, but the paralysed limb fails to move."

But in so dismissing the final stage of volition preparatory to movement, William James surely has evaded what to us neurologists are the real issues. Kinnier Wilson (1928) never allowed us to forget the variability of voluntariness. An individual descending a flight of stairs in the dark illustrated, for him, a transition from a very cautious deliberate motor act to one of facility, speed and little or no willed effort. Much earlier, Hughlings Jackson (1931) spoke of actions which were "most voluntary" and "least voluntary"; "most automatic" and "least automatic", and he even applied these terms to anatomical as well as physiological ideas, segments as well as movements. Indeed he was chary of using the word "voluntary" at all, pointing out that being a psychological term, it was not the right word to use when speaking of physical processes.

The experience of paralytic states bears out these points. William James' conception of paralysis was too simple. We neurologists know that an individual muscle may be paralysed in one context but not another. After ablation of the motor cortex, Denny-Brown and Botterell (1948) noticed that any weakness was always most apparent in simple isolated purposive movements when the animal was quiet. The weakness was less obvious when the movement was motivated by desire to escape from its captor. It was still less severe when the animal clutched at objects in falling. Often no paralysis at all was to be noted when the limb was used in behaviour motivated by rage or fright. Gellhorn (1953), too, had found that monkeys paralysed from poliomyelitis showed an improved performance under the stress of emotional excitement. It is difficult then to describe these varying sets of circumstances in which some degree of movement occurs, either as being volitional or as taking place outside of volition.

An ontogenetic study of willed movements has too long been the perquisite of psychologists rather than neurologists, starting with the spontaneous motor activity of premature, newborn infants. The mass-movements of a young baby can scarcely be deemed volitional, for they are amorphous, bilateral and widespread, irregular in appearance and in rate, unco-ordinated and purposeless. Rather should they be regarded as the precursors of voluntary movements. Later in infancy, a gradual process of increasing economy and efficiency takes place; fragments of this disarray of movement become isolated, and gradually achieve a measure of control. Thus a movement becomes capable of being initiated, or of being inhibited, at will.

According to Ferrier (1886) this is brought about by an organic nexus which develops between special sensory centres and motor centres, and so permits the gratification of a specific desire by way of an individual action. Ferrier followed Bain (1864) in associating a factor of attention with centres of inhibition causing acts of volition to lose their impulsive character and acquire an aspect of deliberation. Whether this is a process of aggregating synaptic connexions or whether it is one of "individuation" or inhibition of accessory responses, is, according to McGraw (1933) not yet known. In Jackson's parlance this is a process of evolution from the most general to the most special; a continual and gradual "adding on" of new organization, that is elements which are most special: but at the same time there operates a steady process of "keeping down" of lower nervous arrangements.

Here then are the beginnings of a deliberate volitional movement; all associated muscular activity becomes regulated, though not at a conscious level, so as to form a harmony of movement, of which the prime movers constitute the melody. Thus a synergic unit is achieved. Of this, only the prime movers carry out the deliberate, volitional, conscious part of the act, the other components of the movement taking place at varying levels of unawareness. "The will", as Kinnier Wilson put it, "makes a subjective but not an objective contribution to movement". Our most detailed ontogenetic information concerns the development in a normal infant of the act of reaching for an object, picking it up and conveying it from one place to another. Using the language of industrial psychology, we can speak of this movement-complex as comprising four functional units or "therbligs", *viz.* (1) transport empty; (2) grasp; (3) transport; and (4) release load. In acquiring such a simple motor achievement, the infant passes through many laborious stages, an apprenticeship of trial and error, with a gradual, though uneven, progress over a period of some sixty weeks. Should mental development be slow, or should states of spasticity exist, these various motor accomplishments are achieved with undue tardiness; the learning process may even be arrested at any stage.

After the normal child has learned how to carry out a modest stock of voluntary movements, he continues to acquire adroitness in certain complex movements over the course of the next few decades, depending upon the opportunities afforded by choice and vocation. Some of these skilled movements may reach such a pitch of efficiency as to constitute "expert movements". Although there is now a considerable literature upon the psychology of manual skill and upon motion-and-time study, the neurological side has been neglected. This is a pity, because neurologists and psychologists both would benefit from co-operation in this field.

Briefly it can be said that by some process of training—whether this be practice or learning—a particular set of movements—simple or complex—can eventually be effected with a high degree of efficiency. A skilled movement is not necessarily a delicate one, any more than an unskilled movement is of necessity a coarse movement. On the contrary, some skilled and even expert actions are comparatively coarse, as for instance, in tennis and cricket. What precisely are the criteria, objective and subjective, which distinguish a skilled from an unskilled movement?

(1) A skilled movement implies that the particular motor act has probably been performed on many previous occasions. The number of practice efforts probably bears an inverse ratio to the degree of "m" or the alleged factor of innate mechanical aptitude.

(2) A skilled movement entails less voluntary effort than a volitional movement. Thus a woman may be able to continue knitting while reading a book; a typist's thoughts may be miles away; a pianist may carry on an animated conversation. Skilled movements, indeed, can often be regarded as automatic movements. But the converse is not true: automatic movements are not necessarily skilled movements. Some skilled movements never become "automatic", however often repeated. Thus the skills shown by jugglers and acrobats always demand vigilance, perhaps because proximal

muscles are concerned rather than digital manipulations: and perhaps because the melody of movement is not fixed or predetermined, but is impromptu, and depends upon a play of varying afferent factors. Expressed otherwise, it belongs to the pursuit type of adjustment.

( ) Skilled movements are as a rule carried out with greater speed than volitional movements, especially when complex in nature. The factors which ordinarily determine the rate of a given volitional movement are obscure. At the command "touch your nose" or "tap the table", a subject will perform these acts at a rate varying between a normal minimum and a normal maximum. This applies when the average inexperienced person first executes a complicated manœuvre; in this respect the expert is much faster. In certain disease-states, movements—whether simple or skilled—are performed much too slowly, even though the strength be unimpaired. This is a matter which cannot, however, be discussed here.

(4) There is far greater economy of movement in the case of the expert, with a more limited range of activity of the prime-movers. Skilled movement is restricted to the needs of the situation, but an unskilled voluntary act may occupy a greater spatial extent.

(5) Skilled movements tend to dispense with associated movements, and with movements of co-operation.

(6) Skilled movements are performed with a less forceful contraction of the prime-movers, than are voluntary unskilled movements: that is, with less expenditure of energy.

(7) Skilled movements—like involuntary movements—are less fatiguing.

Dexterity can be transferred to the unpractised hand. Thus a right-handed person who has learned a skill with his preferred hand, finds it possible to perform that act with the left hand more efficiently than a beginner, even though he may never before have attemped that movement with his left hand. Furthermore, a short period of practice will allow an adept to achieve with his left hand the same high degree of skill as with the right. This fact explains the ability of a hemiplegic artist to continue painting with his other hand without detriment to his technique. Dexterity can also be consigned from one body-segment to another, though with a lesser degree of correlation. Thus an ataxic painter can soon learn to manipulate a brush held between his toes or his teeth. These examples of shifting distribution of the instrumentalities of a learned skill should be of much interest to neurophysiologists.

Whether skilfulness achieved in one complex act can be transmitted to another is less certain, and is still argued. Does an ability to play a violin assist in the technique of typewriting or of embroidery? Perhaps the answer depends upon the nature of the training which led up to the original manual skill. Should this have resulted from a matter of practice, i.e. simple prolonged repetition, it is doubtful whether dexterity can facilitate the development of other skills. But if the dexterity has stemmed from a process of learning, then it probably can be transferred to other complex movements, as industrial experience shows. Learning (in the strict sense) differs from practice in that it educates the apprentice in the fundamentals of motor efficiency, which stand in good stead whatever new task is concerned. Then, of course, there is Thorndike's (1931) suggestion, that practice in one skill may favourably affect practice in another skill because both possess some elements in common.

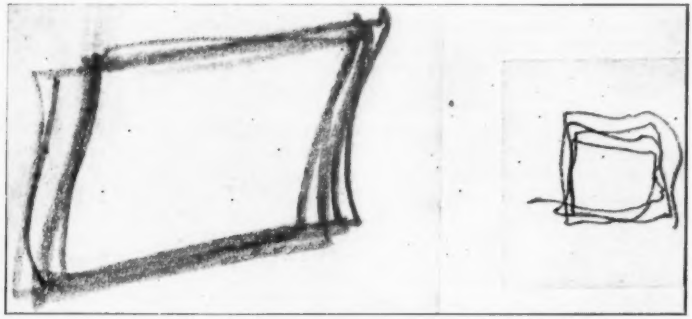
Knowledge of the difference between dexterous and unskilled accomplishments can be incorporated within the systems of learning, and improve industrial output. Various "principles of motion economy" have been enunciated: as by the Gilbreths (1919) and by Barnes (1949). Shaw's (1952) five principles are the simplest. According to Shaw, detailed movements should be (1) simultaneous, (2) symmetrical, and (3) natural.[1] The path of whole movement-cycles ought to be (4) rhythmical, and (5) habitual. By simultaneity is meant that movements should begin and end together; while symmetrical movements are oriented to an imaginary line through the centre of the body. It is important to avoid monotony, by breaking up the rhythm of repetitive small-range movements with occasional larger movements. There are many levels of learning up to a study of fundamental principles. Important in apprenticeship is the formation of correct habits of movements; the means of breaking old and inefficient habits. Learning curves illustrate the rate of progress. At first the advance is rapid: then it slows: indeed progression may halt and the pupil may even fall back a little. These constitute the *plateaux* of the learning curve. Whether these plateaux represent periods of increasing integration, or whether they indicate the pauses between successive methods of learning, is uncertain. How best to allocate the available time into working-periods and rest-periods is another matter for expert advice.

Are special cerebral structures concerned with the dexterous execution of complicated motor skills? Or are they "represented"—if that term can be excused—in the motor cortex in the same way as other movements? Contradictory statements upon this matter have come not only from different writers, but also from the same writer on different occasions. That cerebellar, striato-pallidal, and proprioceptive influences occur in both kinds of movement cannot be gainsaid. But skilled volitional movements as opposed to unpractised volitional acts possibly entail a wider extent of cortical activity

[1]Aristotle insisted that "natural" movements were less tiring than those which were awkward or associated with faulty posture. Thus in his *Problemata*, Chap. V, we read "why is it more difficult to apply prolonged friction oneself to the left leg than to the right? Is it because though our right is the side which is capable of exertion, yet the rubbing of the left leg, since it involves a distorted attitude, is unnatural, and anything which is unnatural is difficult".

in "elaboration fields" lying both in front of and behind the motor area, and also no doubt in homologous but contralateral parts.

Clinical experience shows that skilled movements (that is dexterous, practised movements) are highly vulnerable to the brunt of cerebral disease at all levels: pyramidal, cerebellar, pallidal, and proprioceptive. This observation supports the idea that the integrity of all these mechanisms is needed for the correct and effortless execution of acquired skilled movements. In view of what we know about the ontogenetic development of movements, it would be interesting to enquire whether the pattern of dissolution resulting from disease in any way recapitulates the stages of development, especially when the disease is of a steadily progressive order (*see* Figs. 1 and 2).

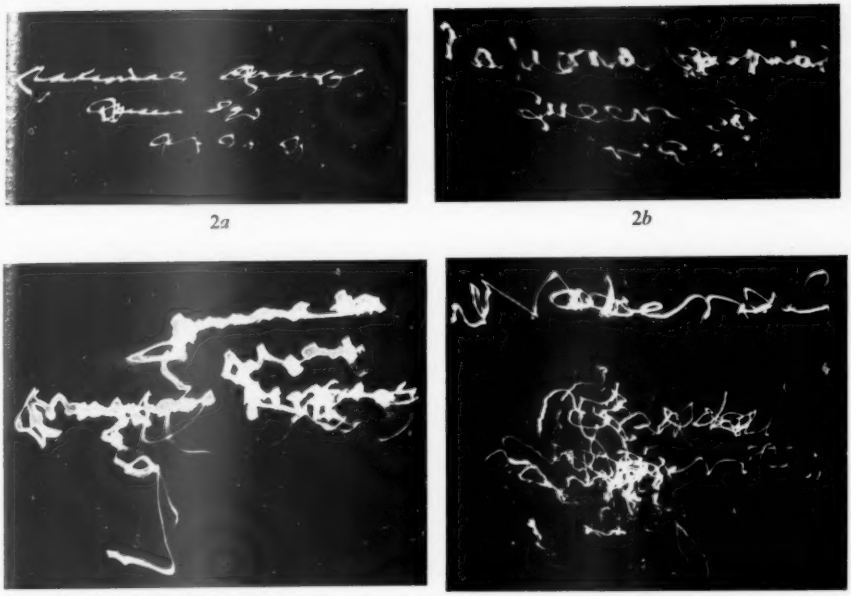


a *b*

FIG. 1.—Drawing squares as rapidly as possible for eight seconds. (*a*) Control. (*b*) Patient with primary cerebellar degeneration.

Neurologists also encounter a dilapidation of skilled movements, apparently in specific and isolated form, in other morbid conditions. We refer, in the first place, to the *apraxias*. Here, by definition, occurs a defect in the execution of a movement, though motor power, sensibility and co-ordination are seemingly intact. Of course, there are many patterns of apraxia, and more orderly thinking upon this subject is much overdue. One difficulty lies in the patient's inconsistencies in performance, for he may do well one moment and fail badly the next. On the whole, the more automatic the movement the better will it be performed. To some extent then, apraxia is more a disorder of simple volitional movement than of skilled movements—in so far as the latter can be regarded as automatic, and independent of deliberate and conscious control. (I shun the term "attention" here, as being still a contentious subject among psychologists.) But if apraxia is not the best example of a specific disorder of skilled movements, we can point to another condition which more adequately applies, namely the various *craft-palsies*. These essentially constitute a specific impairment of educated motor skills. The disability cannot be fully overcome either by an effort of deliberate concentration (i.e. by converting them into highly volitional movements); nor by downgrading them into "more automatic" movements. Such craft-palsies exclusively affect—in the early stages at least—a particular complex movement which has achieved dexterity through tedious practice, and they spare all other volitional movements whether skilled or unskilled, and furthermore, all movements belonging to Jackson's category of "least voluntary" and "most automatic". To look upon the craft-palsies as problems in psychopathology has not proved satisfying even to psychiatrists. On the contrary, the craft-palsies are fascinating problems to the physiological-psychologist and also to the neurologist. The type of error made, for example, by the victim of writer's cramp, is reminiscent of many of the distinctions between an unpractised complex movement, and one which is deft and practised (*see* Figs. 3 and 4).

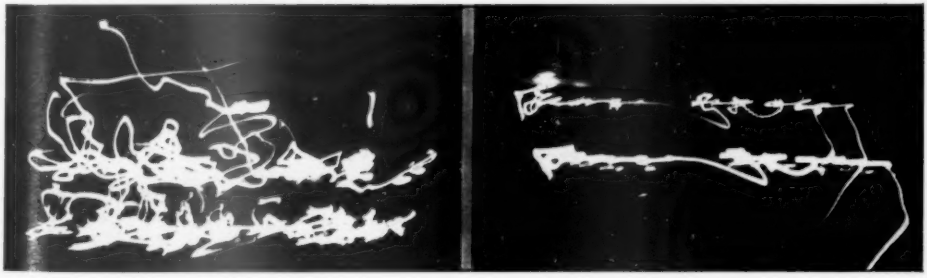
Brief mention should be made of an unfamiliar type of skilled movement. I refer not to practised movements of extreme complexity, but rather the reverse. Under the term *skilled minimal movements* one can allude to those unusual tricks of motility whereby a single muscle, or a part only of a single muscle, can be made voluntarily to contract. I deliberately avoid speaking of "discrete movement". Typically the muscle concerned is not one which participates in ordinary manipulations, but is a proximally situated muscle, or still more characteristic, part of the muscular investment of the trunk. By dint of practice, some few are able at will to contract and relax the muscles attached to the auricle of the ear; or to move one-half of the frontalis muscle, or a segment of one pectoralis major, or deltoid, or rectus abdominis. Students of the Orient will recall the startling repertoire of unexpected minimal muscle-movements which can be brought within the category of volitional movements. The mechanism of production of these movements entails a sort of practised fractionation, analogous to the infant's increasing power of controlling isolated fragments out of a mass of movements.



2*a* 2*b*

2*c* 2*d*

FIG. 2.—Cyclegram of the movements of the index finger during the act of writing "National Hospital Queen Sq., W.C.1." (*a*) Normal right-handed subject using right hand. Time taken 13 seconds. (*b*) Normal right-handed subject using left hand. Time taken 28 seconds. (*c*) Patient with Huntington's chorea using right hand. Time taken 54 seconds. (*d*) Patient with Huntington's chorea using left hand. Time taken more than 85 seconds.



3*a* 3*b*

FIG. 3.—Writer's cramp. Cyclegrams of the movements of the index, middle and ring fingers of the right hand during the act of writing. (*a*) Patient with writer's cramp. Time taken 14 seconds. (*b*) Normal subject. Time taken 6 seconds.

One may also mention as another fitting theme for neurological attention, the subclinical muscular contraction, or perhaps only action potentials, which can be detected in muscle groups in circumstances of mere imagery of movement; or during the mental state of recalling muscular acts, or of matters which would involve such a motor concomitant. The importance of these observations, which have been made for nearly a century and which were predicted by Jackson and studied by Lotze, Bain Ferrier, Golla (1921), and Jacobson (1932), is a fundamental one in psychology for they throw light upon the nature of thinking processes, and suggest that the activities of thought are not confined to closed circuits within the brain. These "shadow movements" do not actually fall within the ranks of volitional movements, though they may be looked upon as the expression of the recollection of volitional movements.

Lastly, in a discussion upon volitional activity, a neurologist must decide where precisely b long mimic movements of gesture. And where, too, should we place the tics, especially the more elab rate, quasi-purposeful habit spasms? If we subscribe to the doctrine that there are levels of voluntar ess, then we cannot rightly exclude either gesticulation or the tics, mindful of the definition mad by Meige and Feindel (1907) . . . "a tic is a co-ordinated purposive act, provoked in the first instan by some external cause or by an idea; repetition leads to its becoming habitual and finally t its involuntary reproduction without cause and for no purpose, at the same time as its form, inten sity, and frequency are exaggerated; it thus assumes the characters of a convulsive movement, inoppo une and excessive; its suppression associated with malaise. The effect of distraction or of volit nal effort is to diminish its activity; in sleep it disappears. It occurs in predisposed individuals, who usually show other indications of mental instability. . . ." Like ordinary volitional movements tics are fully co-ordinated actions, preceded by a desire for their execution, and succeeded by a fe ling of satisfaction. At the same time a tic is an inapposite and incongruous act which may in certain circumstances even prove harmful. Hence it has been well termed a parasite function. A tic, like a gesture, is a kind of voluntary movement, low in the ladder of volition.

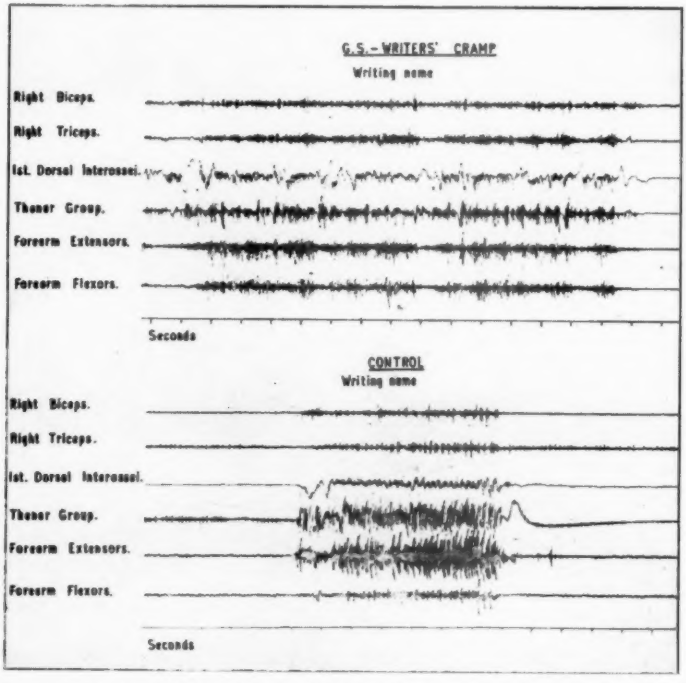


FIG. 4.—Electromyograph taken during the act of writing, in the case of a normal subject (below) and a patient with writer's cramp (above). (Courtesy of Dr. W. A. Cobb.)

SUMMARY

My purpose has been to stress the fact that volitional movement is a far more complex idea than would appear at sight. Although in the past efforts have been made to align electrically induced motion with volitional movement, one of my objects would be to emphasize the dangers of such an analogy. As to the important contributions of experimental physiology a quotation offers itself as apt.

". . . Our survey must be circumspect for several reasons. By use of such methods as we are employing, artificial excitation, and so on, and under such observations as these allow, namely the initiation under narcosis of muscular movements or the recording of their immediate defects from normal movement, little light is given in regard to much that goes on in an organ whose chief function is mentality itself. Our expectation must be modest, for modest must be the achievement reached by such means in a problem of such a nature . . ." (Sherrington, 194 .

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Dr. J. A. V. Bates: The concept of "voluntary movement" was taken from mediæval philosophy and carried into clinical neurology. The words "voluntary movement" mean the same today as they meant in the seventeenth century, i.e. "movement which proceedeth from the Will" (Hobbes, Leviathan, 1651); "volitional movement" is a more recent synonym. Clinical neurologists gave the faculty of voluntary movement a cortical locus about 150 years ago, and since that time the consequences of cortical injury have been referred to as a "paralysis of voluntary movement". Ferrier, eighty years ago, claimed that the results of stimulation and ablation demonstrated the precise "cortical centres for voluntary movement"; three years later (1876) it was shown that the pyramidal tract originated at least in part from the precentral gyrus, and a route by which "voluntary impulses" could reach the spinal cord was seen to be available. Ferrier's interpretation of his results was accepted by most of his contemporaries. The precentral gyrus and the cortico-spinal tract were identified as the system mediating voluntary movements and this idea has had a strong appeal to neurologists and is still the current textbook teaching. In Ferrier's original scheme the basal ganglia were an integral part of the voluntary motor system, but when it was discovered that they were by-passed by the cortico-spinal tract they, together with other non-pyramidal efferent pathways and their respective stations of origin, became an "extrapyramidal system" mediating all non-voluntary movements. The scheme of an extrapyramidal system mediating involuntary movements was a necessary and inevitable consequence of the scheme of a pyramidal system mediating voluntary movements.

There are adequate grounds for holding that the original identification of the pyramidal system with the concept of voluntary movement was based on an over-enthusiastic interpretation by Ferrier of the type of data offered by physiological experiment and clinical observation. In the first place, the movements evoked by cortical stimulation have not the correspondence with skilled or purposive movements which Ferrier claimed for them; secondly, following cortical destruction, the subject is more adequately described as suffering a depression of certain motor abilities, than as suffering from a paralysis of voluntary movement. This is an important distinction.

In addition to these original errors of inference, recent observations have supplied evidence which further weakens the foundations of Ferrier's scheme. These observations are as follows:

(1) The basal ganglia exposed after hemispherectomy in man are inexcitable to stimuli which excite the motor cortex and the cortico-spinal tracts and are therefore not motor in the sense that the precentral gyrus is motor. Practically complete ablation of these ganglia in one case has not produced any further symptoms. There is no experimental support for an involuntary motor system associated with these ganglia, and both voluntary and involuntary movements in the neurologist's terminology require the integrity of the cortico-spinal system. This has long been appreciated from work on animals. (2) Observations following stimulation of the white matter in man give no reason for supposing that the grey matter of the precentral gyrus contributes to the fineness or complexity of an individual response. Judging from the results of stimulation, there are two principal phenomena dependent on the presence of this grey matter. They are secondary facilitation, and the clonic "after-discharge" movements as seen in focal epileptic seizures. There is no evidence to support the idea that fine gradation of movement is a property of the grey matter of the precentral gyrus. (3) The ability to make a movement to command (e.g. to close the fist) can survive hemispherectomy even when an apparently identical movement has been produced by cortical stimulation of the diseased hemisphere before excision. This is not a "recovery phenomenon", the movement can be made immediately consciousness is regained. Thus, a positive result from stimulation is not *invariably* linked to an immediate loss of the corresponding movement after ablation.

These facts, together with the classical work of Lashley (1924), Jacobsen (1934), Denny-Brown and Botterell (1948) and others on the retention of motor skills following ablation of the precentral region in animals, suggest that the association of the concept of voluntary movement with the pyramidal system, and *per contra* the association of involuntary movement with an "extrapyramidal" system, is a false association of incompatible concepts, founded on an erroneous interpretation of data, and for this reason is impeding a proper understanding of the physiology of the motor system.

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Professor E. G. T. Liddell: Volitional movement is studied from different angles by different disciplines. The physiologist, while recognizing something which can be called "volition", has little or nothing to say about it. His efforts are bent on a low level of action: the execution of movement. "Volition" in animals, and in his animals often anæsthetized at that, is beyond his ken. Any movements of limbs which he may study may be superficially similar to volitional movements, though in reality they are only fragmentary caricatures of volitional movements. Sure enough, they are movements of limbs which are moved by volition in the normal animal, but that does not justify his experimental movements being called "volitional movements".

At the executive level where physiologists have laboured for so long, there is the great cortico-spinal pathway debouching upon spinal motoneurones. There is also within reach of investigators nowadays the vast sensory control, both cortical and subcortical, which presides over muscular movement. In addition, at spinal level, there is the control of spinal motoneurones by themselves which has lately been revealed with enchanting clarity. This proprioceptive system has long been recognized. Sixty years ago, Sherrington (1894) pointed out that about one-third of the fibres in the nerve "to" skeletal muscle were, in fact, "from" the muscle to the reflex centres. Only the remaining two-thirds really were "to" the muscle, being "efferent" fibres. But now we know that one-third of the two-thirds, two-ninths of the whole nerve, are small motor fibres, 3–8μ in diameter, the so-called γ fibres (Hunt, 1951; Hunt and Kuffler, 1951). They are almost constantly transmitting impulses to the small intrafusal muscle fibres of the muscle-spindle. Thus they are tensioning and alerting the sensory corkscrew in the muscle-spindle, the well-known annulo-spiral ending of Ruffini. 10–60 impulses a second may thus pass to the intrafusal muscle fibres. Nothing is added to skeletal muscle tension by these intrafusal fibres but the sensory ending of Ruffini is brought into action by the intrafusal fibres and it sends messages ("feeds back") to the motoneurones of its own muscles. These in-going "afferent" impulses travel express by large fibres 10–20 μ in diameter at a rate of 60–125 metres a second. More than that, the expressness of their journey is aided by the pathway being mono-synaptic, so that they reach the motoneurone with the smallest possible delay in a total time of about 5 msec. Herein lies the nervous mechanism of the spindle action, elicited in two ways (i) by increased tension from its own intrafusal fibres, or, of course, (ii) by increased tension from outside the muscle applied through the tendon, as it is in any tendon reflex or stretch reflex, or by both of these means (i) and (ii) at one time.

It is well known and often repeated that "posture follows movement like a shadow" and that "every movement begins in posture and ends in posture". So in the cat, when the pinna of the ear is titillated to elicit the scratch reflex, the physiologist finds nowadays by his refined method of experiment that the small γ nerve fibres of the lower limb carry an increased rate of discharge: the intrafusal muscle fibres contract more: the sensory corkscrew of Ruffini is more alerted: in turn the spinal motoneurones are alerted: and the posture of the lower limb is thus got ready to pass into the movements of the scratch reflex and scratch the titillated pinna (Granit *et al.*, 1952). Some preliminary experiments in the cat have shown also that many structures well above spinal level can, through the γ nerve fibres, influence the intrafusal muscle fibres and alert a muscle's posture—such structures as the anterior lobe of the cerebellum, the diencephalic reticular formation, the motor cortex, the habenula, and the medial head of the caudate nucleus. Deceleration of spindle activity was obtained from the bulbo-reticular system and from the anterior lobe of the cerebellum in decerebrate as well as in Dial-chloralose animals (Granit and Kaada, 1952). The population of muscle-spindles is quite high. In a medium-sized cat the slender little soleus muscle may weigh about 1½ grammes and contain 45 muscle spindles. A cat's gastrocnemius had a population in one example of 56 spindles, and tibialis anterior 57 (Hagbarth and Wohlfart, 1952). A muscle of comparable size in man is the inferior rectus of the eye in which were found 47 spindles (Cooper and Daniel, 1949). Some significant observations have been made in stretch impulses in the anæsthetized goat, from the jaw muscles and extrinsic ocular muscles. Strong messages were detected in the mesencephalic nucleus of the V nerve and faintly in the cerebral cortex. That stretch messages should thus be found in the cerebral cortex is surely one of the most pregnant contributions to the study of conscious proprioceptive sensation in recent times. The messages reached the cortex after long delay (200 msec) but that they were detected there at all is a most notable discovery (Cooper *et al.*, 1953).

I have said nothing of the muscle's own automatic inhibitory control of itself by the Golgi tendon organs. Their simpler downright action in spinal motoneurones has been made clear enough by modern methods, skilfully and carefully used. They can bring their inhibition into play 5 msec after a single excitatory wave from the annulo-spiral ending of Ruffini has begun and continue it for another 25 msec, ultimately entirely abolishing the excitation (Granit, 1950).

I have written at some length about this automatic regulatory system of the lower motor neurone because the exquisite beauty of its function must be borne in mind when we come to consider the upper motor neurone. The upper motor neurone has, of course, long been under scrutiny. One of the great histological surprises of recent times has been to find how few large axones coming presumably from large cortical cells, indeed less than 2% of all the fibres, can be counted in one human pyramid at medullary level and how large is the proportion of small fibres, 98% of a pyramid (Lassek, 1942). The old controversy, now appearing to be settled, at any rate for the time being, is the place of termination of its fibres among the spinal neurones. In a former generation, Schäfer (1899) was loud in asserting that the fibres ended in the posterior horn of grey matter. That view has been reaffirmed by modern histological methods (Szentagothai-Schimert, 1941), as well as by electrical methods (Lloyd, 1941). Unlike the muscle's self-excitation from its own annulo-spiral endings, the pyramidal messages do not go by express paths to the spinal motoneurones. The first impulse in a train of pyramidal messages seems to get lost in the external basilar region at the base of the posterior horn among whose cells it dawdles for 40–45 msec. Later impulses may pass the anterior horn cells much more quickly, in 1 msec, apparently by the opening up of progressively shorter functional pathways. By a slightly different kind of experiment, it was made clear that messages down the pyramidal tract must be repetitive and that by repetition they increase the alertness of spinal motoneurones (i.e. "facilitate" them) though without shortening their latency of action. These observations on the need for repetition of pyramidal impulses raise the question of the nature of discharge from cortical cells. If their discharge has to be repetitive to be effective, how is that repetition brought about? To be sure, there are cells enough and pathways enough in the cortex to give, at any rate, localized circuses for impulses, localized within one or a few cortical units, but there is not yet much direct evidence to hand on that topic. One can only speculate at present. We need closer, finer analysis of the excitation of cortical cells. The classical methods of cortical stimulation leave a good deal to be desired. The depth of anæsthesia cannot be maintained with certainty from moment to moment when volatile anæsthetics are used. The electrodes themselves, even when held mechanically and not manually, are uncertain in effect because electricity, coursing through some millimetres of saline-filled non-homogeneous tissue of the brain certainly could follow capriciously a variable path. With an oft-repeated train of stimuli, as in faradization, there is evidence that, besides the main motor cells being activated, there are subsidiary inhibitory cells activated simultaneously to produce a sort of "latent inhibition". This results in a rebound contraction after the end of the electrization, which subsides into a complete inhibition of contraction as shown both myographically and electrically. This kind of movement is therefore a long way from "volitional" movement. The brain is anæsthetized, too, and if its surface under the electrodes be inspected with a microscope, there is seen a profusion of tiny bubbles of hydrogen and oxygen rising from the surface (Denny-Brown, 1927) and alterations of blood flow in the capillaries: all factors which should lead to modest appraisal of the experiments. There is still another factor inviting caution and that is the long latency before the muscle begins to contract at all. The latency is much longer than that of a voluntary movement. These features of long latency, of latent inhibition and of rebound are all often found also in the classical crossed extensor response in which, because a "mixed" afferent is often used, there is often mixed excitation and inhibition on the crossed side, with excitation winning most of the time, while on the side of the afferent nerve, the prepotent flexor reflex is in action, because there flexion wins over any latent tendency to extension. Muscles responding to cortical stimulation or to crossed nerve stimulation give very similar myograms. So safe conclusions from experiments on cortical stimulation, especially when faradic stimuli are used, follow only on cautious assaying of experimental procedures used. In experiments of this kind, the physiologist has not been studying "volitional" movement but the responses of part of the mechanism when it is treated abnormally. Muscular co-ordination in normal life and the posturing of the rest of the body while one part of it performs a muscular act, are fit subjects for physiological investigation but they are only glimpsed as yet.

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[*February* 4, 1954]

Clinical Aspects of Carotid Thrombosis

By G. E. SMYTH, M.D., F.R.C.P.

Manchester

My observations are based on a study of 19 cases collected from the records of the Manchester Royal Infirmary. All have occurred within the last five years and for access to some of the cases I am indebted to the kindness of my colleagues. I have no doubt that this number does not give us a true notion of the incidence of the condition, for, on the whole, we have tended to limit the use of angiography to cases of suspected cerebral tumour and when the lesion has appeared to be clearly vascular in nature we have been somewhat diffident about the propriety of carrying out angiography. The increasing use of angiography will doubtless before long make this a common, if not indeed a fashionable, diagnosis. In this brief study I shall not concern myself with pathological or radiological details, but will limit my remarks to an analysis of the clinical features.

OF the 19 cases all but one occurred in the fifth and subsequent decades. In 8 cases the disease showed itself by an abrupt and severe hemiplegia. In the other 11 the onset was either gradual or remittent. Repeated or slowly increasing monoplegia was a feature of 6 of these 11 cases and in 3 there were recurrent attacks of dysphasia. The 2 remaining cases in this group showed curious sensory disorders in one hand, recalling, in fact, a peripheral nerve lesion, and the true diagnosis was only revealed when a definite hemiplegia finally made its appearance. In the great majority of cases paralysis greatly predominated over sensory loss. Hemianopia occurred in only one case. It is plain that in many respects and, so far as this study goes, in regard to the age incidence, this condition offers few striking differences from classical cerebral thrombosis. That the diagnostic difficulties are great is further shown by the fact that in no less than 8 of these 19 cases neoplasm was considered as the most likely diagnosis.

In the present state of knowledge it is probably impossible to arrive at a correct diagnosis without the aid of angiography. Our suspicions should, I think, be aroused when we are confronted with attacks of recurrent paresis or dysarthria. These recurrent or relapsing symptoms have, on several occasions, been observed to occur over as long a period as two years. In this series we did not obtain any help from the degree of pulsation in the carotid vessels and in fact in only 3 instances was it thought that the pulsation was significantly diminished on the affected side. Up to the present, transitory loss of vision in the ipsilateral eye has not been encountered and, in common with most observers, we have not met with permanent ipsilateral blindness following the final attack of thrombosis.

At the present time the differentiation of this condition from ordinary cerebral thrombosis is probably of academic interest, but the time may come when its recognition will have practical importance.

Thrombosis of the Carotid Artery

By J. L. G. THOMSON, M.D., M.R.C.P., D.M.R.D.

National Hospital, Queen Square, London

To obtain some idea of the frequency of carotid thrombosis in cerebral angiography the last 1,800 angiograms performed at the National Hospital, Queen Square, were reviewed. Among these were found 22 cases of carotid thrombosis, giving a percentage incidence of about 1·2.

Analysing the positions at which the block was demonstrated, it was shown that one was in the common carotid artery, 16 were either at the origin of the internal carotid artery or within its proximal 3 cm., and the remaining 5 cases were in the carotid siphon.

The clinical diagnosis prior to angiography was also noted and it was found to be correct in 7 of the cases. In 2 other cases an aneurysm on the carotid siphon had been diagnosed, and after the block had been shown, it was assumed that the aneurysm had probably thrombosed and that this process had spread to involve the main artery. Both patients had a palsy of the III and VI cranial nerves on the same side as the thrombosis, and one showed a ring shadow on the plain X-ray of the skull, which probably represented the calcified aneurysm.

6 other cases were diagnosed as having a cerebral tumour, 4 as a cerebral vascular accident, 2 as a cerebral angioma, and in 1 no diagnosis was made.

In one of these cases, autopsy examination revealed a carcinoma of the bronchus with cerebral secondaries as well as a bacterial endocarditis; so it is possible that other cases may have had a different underlying pathology, with the carotid thrombosis as a complication.

Examples of the cerebral angiograms in these 22 cases were then shown by lantern slides. 3 are perhaps worth noting:

Firstly, a thrombosis in the left common carotid artery (Fig. 1), which shows an absent filling

FIG. 1.—Thrombosis of the common carotid artery.

FIG. 2.—Thrombosis of the internal carotid artery showing collateral circulation.

FIG. 3*a*.

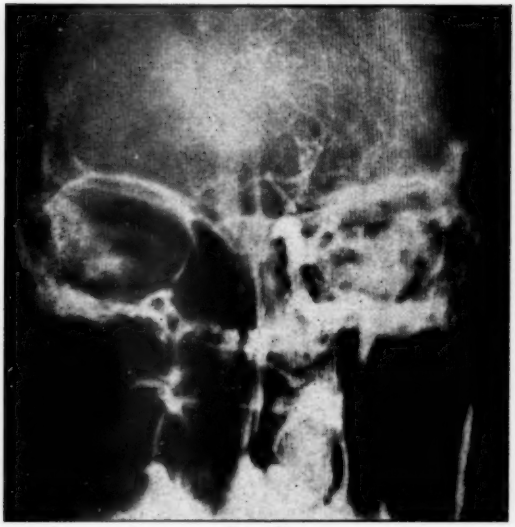

FIG. 3*b*.

FIG. 3*a* and *b*.—Thrombosis of the carotid siphon. The ophthalmic, posterior cerebral and anterior choroidal arteries are outlined.

in the external and internal carotid arteries, although good retrograde filling down the common carotid artery can be seen. This appearance was obtained from a 49-year-old female, who had been developing a progressive right hemiparesis and dysphasia for two years. Her blood pressure was 135/90.

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Secondly, an example of the collateral circulation, which is known to exist between the external and internal carotid arteries. Fig. 2 shows contrast medium passing from the maxillary artery to the orbit, and from there the ophthalmic artery and carotid siphon are outlined. A by-pass round the blocked internal carotid artery can thus exist or become established.

The third example (Fig. 3 *a, b*) is of a block in the carotid siphon, in this case just beyond the origin of the anterior choroidal artery. The ophthalmic and posterior cerebral arteries are also outlined.

Emphasis was also laid on the importance of obtaining films both of the stump of the blocked artery and also of the needle point, before making a confident diagnosis of carotid thrombosis. Confusion can otherwise easily arise.

Some Aspects of the Pathology of Thrombosis of the Internal Carotid Artery

By Brian Turner, M.B., B.S.

Histology Laboratory, The National Hospital, Queen Square, Rockefeller Travelling Fellow from the University of Sydney, Australia

Thrombosis of the internal carotid artery is more common than is usually expected, for Hultquist (1942), investigating the carotid arterial system in 1,300 routine autopsies, found a thrombosed internal carotid artery in 4·4% of the cases.

In the 4 cases under consideration the thrombosed artery was the site of marked atheromatous change as were the other arteries in the body.

Atherosclerotic deposits are often found in the internal carotid artery (Dow, 1925; Keele, 1933) and they are usually situated in the region of the carotid sinus and in the carotid siphon which are also the most frequent sites for thrombosis (Hultquist, 1942).

In all the present cases the initial site for thrombus formation was in the region of the carotid sinus. In one there was also old thrombus in the carotid siphon.

The frequency of demonstrable cerebral complications varies with the site and extent of thrombus formation. With thrombosis at the proximal end of the artery Hultquist found cerebral lesions in one-third of the cases. With thrombosis at the more distal level the incidence was much higher.

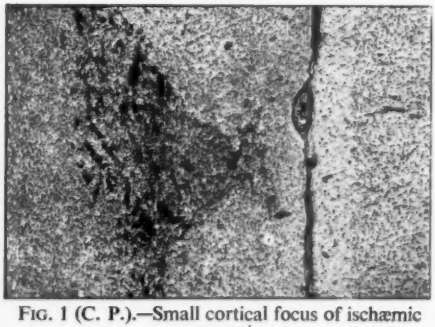

Fig. 1 (C. P.).—Small cortical focus of ischæmic necrosis.

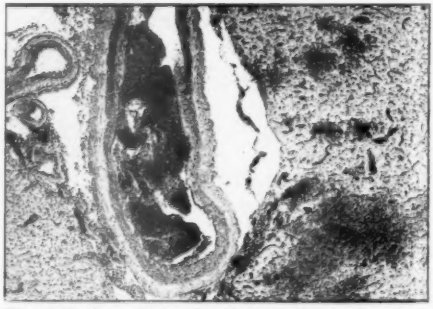

Fig. 3 (C. P.).—Convoluted mass of thrombus in a pial artery. Adjacent brain shows recent ischæmic necrosis.

In the present cases the cerebral changes ranged from ischæmic damage to the cortical neurones to foci of ischæmic necrosis of varying age and size.

In one patient (F. S., No. 1484) the internal carotid artery was partially occluded at its lower end by old organized thrombus and beyond this by more recent thrombus which extended as far as the origin of the middle cerebral artery. In the cerebrum there was massive old ischæmic necrosis in the region of distribution of this vessel. The heart showed an old myocardial infarct with an adherent laminated thrombus in the left ventricle. Emboli from the left ventricle may, therefore, have been wholly or partly responsible for both the carotid and the cerebral lesions.

In 2 cases (S. H., No. 37477 and C. P., No. 24452) thrombosis of the internal carotid artery and discrete foci of ischæmic necrosis in the brain were the principal autopsy findings (Fig. 1). The cerebral damage was almost entirely confined to the distribution of the middle cerebral artery on the same side as the thrombosed internal carotid artery (Fig. 2).

In the second of the 2 cases, close to several of the more recent lesions small cerebral arteries were partially blocked by convoluted masses of recent thrombus (Fig. 3). The walls of these arteries showed no evidence of arterial disease.

Other small arteries in this hemisphere were occluded by older organized thrombi.

The appearance of these thrombi was compatible with their being of embolic origin but it is not possible on histological grounds to exclude their being primary thrombi laid down at these points.

It is suggested that masses of thrombus material may have been dislodged from the thrombus in the internal carotid artery and lodged in the smaller cerebral vessels.

The evidence supporting this proposition is, firstly, that the appearance of these masses is compatible with their being of embolic origin; secondly the distribution of the lesions is in accord with the known distribution of emboli entering the carotid circulation; thirdly the

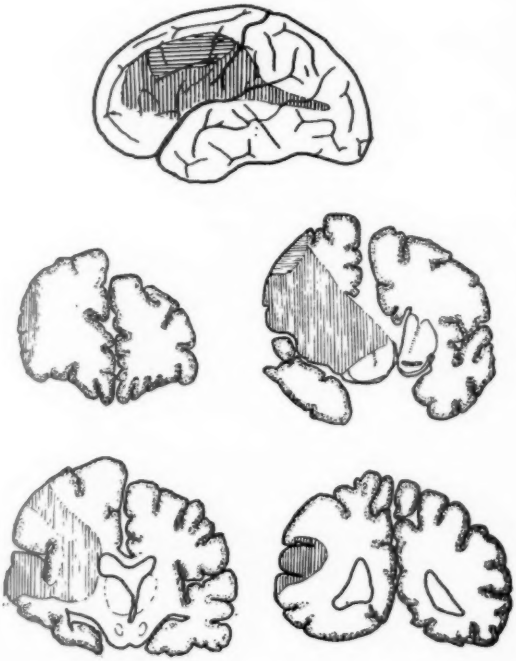

FIG. 2 (C. P.).—Diagram of the extent of the cerebral damage. Horizontal hatching represents recent and vertical hatching older regions of ischæmic necrosis.

discrete focal nature of the lesions with varying degrees of repair and organization; and lastly the episodic clinical course of these cases.

In the fourth case (E. B., No. 36501) there had been a sudden onset of cerebral symptoms and only a short interval between the onset and death. The internal carotid artery was occluded by recent thrombus and in addition the circle of Willis was very atherosclerotic. There was an extensive ischæmic lesion in the region supplied by the middle cerebral artery on the same side as the thrombosed internal carotid artery but, in the sections examined, occluded cerebral vessels were not found. It appeared that the circle of Willis was unable to provide an adequate collateral circulation and accordingly the territory of the middle cerebral artery suffered from an acute ischæmia.

Summary.—It is believed that atherosclerosis is an important factor in the pathogenesis of thrombosis of the internal carotid artery. Emboli arising from the thrombus and lodging in smaller cerebral arteries are a factor in the production of the cerebral complications of some of these cases.

My thanks are due to Sir Charles Symonds, Dr. E. A. Carmichael, Dr. Purdon Martin, Dr. S. P. Meadows and the Medical Committee of the National Hospital, Queen Square, for permission to utilize the case records, and to Dr. W. Blackwood for placing the pathological material at my disposal.

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Cerebral Pathology following Carotid Spasm

By P. O. Yates, M.B., Ch.B.

Department of Neuropathology, University of Manchester

Vasospasm is not a condition which persists after death and is not therefore a fact observable by morbid anatomists. Experimental observations in animals (Kinmonth, 1952) and the experiences of surgeons suggest that prolonged spasm of arteries is not easily produced except by direct trauma to the vessel wall.

An exception to this appears to be the well-known work of Villaret and Cachera (1939) who observed, through cranial windows, the lodgment of solid emboli in vessels of the pia-arachnoid. They noted brisk contractions of the walls not only of the vessel occluded but also of neighbouring arteries. In a few such instances these contractions might be prolonged. Such a phenomenon has often been suggested as the cause of the temporary nature of the neurological disorders which may occur in cases of embolization where the cerebral arteries are otherwise healthy.

A feature of the lesions of atherosclerosis is the presence of thin-walled capillaries and sinusoids in the atheromatous plaques and in the wall of the artery beneath the plaques. Occasionally these rupture, allowing hæmorrhage into the muscular wall of the vessel. I believe such a naturally occurring injury may produce arterial spasm, which, if short-lived, will give rise to fleeting neurological signs and, if prolonged, offers an explanation of infarcted cerebral tissues which at autopsy are seen to be supplied by arteries not thrombosed and only slightly occluded by atheroma.

The damage caused to the carotid artery by angiographic needling which may be insignificant in a healthy vessel may cause a hæmatoma and even extensive dissection of the wall in atheromatous arteries predisposed to such a lesion. Any such lesion may stimulate prolonged spasm of the vessel.

Death within a few hours of angiography sometimes occurs, especially in patients suffering from atherosclerosis, but in these cases time does not allow development of the picture of ischæmic damage which may, in any case, be confused by the primary disease.

A case which I think is significant is that of a man aged 63 who had suffered for two years principally from dysphasia. Left-sided percutaneous carotid angiography was performed apparently satisfactorily, though the films showed that little contrast medium had passed up the internal carotid tree. During the examination he became unconscious and remained so until his death eighteen days later. At autopsy there was slight evidence of bruising of the common carotid artery. Both the common and the internal carotid arteries showed plaques of atheroma but nowhere was there occlusion or any thrombosis. Examination of the brain showed a widespread atherosclerosis of a moderate degree but no obstructed vessels could be found. There

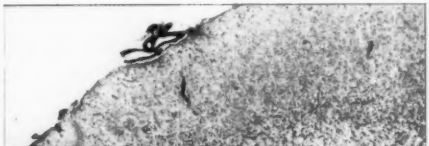

Fig. 1.—Cerebral cortex showing laminar necrosis.

were scattered small areas of softening of varying ages in grey and white matter of both hemispheres. The lesion of particular interest is in the cortex of the left fronto-parietal regions. This lesion consists of necrosis of the third and fourth layers of the cortex extending in places to deeper layers and occasionally joining areas of softening in the underlying white matter (Fig. 1). The damaged zone is occupied by microglial phagocytes and proliferating capillaries and is compatible in age with having been produced at angiography eighteen days previously.

This case presents the picture of a moderately severe cerebral arteriosclerosis with, in addition, ischæmic cortical necrosis predominant in the area of distribution of the left middle cerebral artery. It strongly suggests that severe cerebral damage can be caused by prolonged spasm of the carotid artery and that therefore the clinical and angiographic picture suggesting a diagnosis of thrombosis may be fallacious.

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Dr. J. St. C. Elkington referred to the case of a previously healthy man of 39 who, while working on the railway, was struck on the head by a signal and rendered unconscious for a brief period. Next day he resumed work and continued for three days, when he fainted. On examination at this time he was found to have a right hemiparesis including the face.

On the following day he had recovered and remained well for nine days when he suddenly lost consciousness and was admitted to hospital. He was subcomatose with a right hemiplegia, right hemianopia and aphasia. The fundi were normal. General medical examination was negative. C.S.F. was normal in all respects, including pressure. W.R. negative.

He died five days after admission.

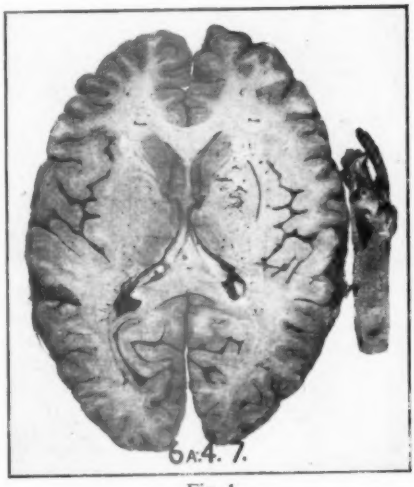

Fig. 1.

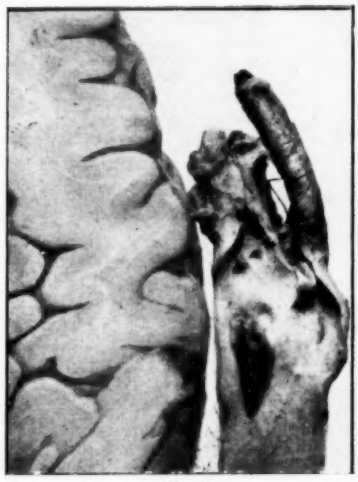

Fig. 2.

FIGS. 1 and 2.—Showing (i) atheromatous ulcer with adherent clot at the termination of the common carotid artery; (ii) thrombosis of the internal carotid artery; and (iii) area of softening in the corresponding cerebral hemisphere.

The specimen shows an area of softening in the left hemisphere (Fig. 1). The middle cerebral artery was thrombosed and full of firm clot. The whole internal carotid artery was similarly occluded. Just proximal to the bifurcation of the common carotid artery was a circumscribed area of atheromatous ulceration to which was attached a firm, dark red blood clot (Fig. 2).

The speaker suggested that his specimen illustrated the part that trauma might play in carotid thrombosis and also the probability that the initial symptoms were embolic in origin.

Mr. Geoffrey Knight said that Dr. George Smyth's paper on the clinical aspects had emphasized the difficulties of diagnosis. To borrow a phrase from Dr. Smyth's paper, it was "worthy of honorable mention" that of the last 4 cases of this condition simulating cerebral tumour which had been admitted to the South-East Metropolitan Regional Neurosurgical Centre, 3 had been diagnosed with confidence by Dr. Simon Behrman prior to the performance of arteriography. Although these cases appeared to

produce a steadily progressive syndrome continuing over a period of months—in one case for eighteen months—careful analysis showed that this syndrome could, in fact, be broken down into a series of successive isolated incidents, each leaving some permanent blemish, which, in the sum, produced an appearance of continuous progression.

One patient had exhibited a syndrome suggestive of a right temporal lobe tumour presenting with a history of progressive personality change followed by the development of a progressive hemiparesis, and investigation had revealed a quadrantic hemianopia. At arteriography in this case excellent filling of the vertebral and basilar arteries was produced by reflux owing to obstruction of the carotid high in the neck.

A second case simulated the evolution of a left temporal lobe tumour with increasing aphasia, paresis of the contralateral arm and epilepsy. In order to account for the successive incidents which led to this progression in the syndrome they had postulated the occurrence of multiple emboli in the small cerebral vessels, a postulate which receives confirmation from Dr. Brian Turner's observations. In order to protect the brain from further damage by embolism they had, in the last 2 cases, gone so far as to tie the internal carotid artery high in the neck, for they believed that if it could be shown that the progressive syndromes were produced by multiple cerebral emboli, surgery had a part to play in protecting the brain from increasing damage.

Professor P. C. P. Cloake said that an interesting problem was the explanation of the long remittent history in some cases of internal carotid thrombosis. The appearance and remission of symptoms might continue for months or even years and attempts to explain this fact had been based on an hypothesis of cerebral embolism or on one of intermittent spasm of cerebral arteries.

Embolism would presumably mean that the carotid thrombosis had already happened when these symptoms began, spasm might precede or follow the carotid thrombosis, but it is difficult to believe that either of these factors alone would be likely to play an important part in producing remitting symptoms over many months, especially since evidence of multiple emboli was usually not found.

It was known that internal carotid blood flow could be arrested in many people without causing any serious effects, the anastomotic circulation becoming at once adequate to supply the necessary blood flow to the deprived cerebral arteries. This anastomotic circulation came from the vertebrals, the opposite internal carotid by way of the circle of Willis and the external carotid artery of the homolateral side mainly via orbital vessels and branches of the ophthalmic artery in which, of course, the flow might be actually reversed if the anastomotic contribution from the external carotid was large.

Though this readjusted circulation might prove adequate to maintain normal cerebral function it was possible that its reserves were much more limited and that relatively minor disorders of systemic circulation might suffice to break down this compensation. When that occurred it was suggested that the symptoms appeared: for example, transient amaurosis of the homolateral eye. Such disturbances might sometimes occur also on the "normal" side.

Dr. W. Ritchie Russell (Oxford) pointed out that thrombosis of the internal carotid artery might cause no symptoms. The arteriographic demonstration of a blockage of the internal carotid artery did not in itself justify conclusions as to what lesions had occurred in the brain.

Histological Demonstrations

Acute Hæmorrhagic Leuco-encephalitis.—Dr. L. Wolman.

(1) **Malignant Neurofibromatosis.** (2) **Myopathy Involving the Pharynx.**—Dr. L. Woodhouse Price.

Section of Obstetrics and Gynæcology

President—Victor Lack, F.R.C.P., F.R.C.S.Ed., F.R.C.O.G.

[*February* 26, 1954]

Reconstruction of an Obliterated Cervical Canal by Transplantation of Endometrium

By Professor M. A. van Bouwdijk Bastiaanse
Amsterdam

In 1947 and 1948 2 patients were referred to the clinic for amenorrhœa, with attacks of pain in the lower abdomen recurring every four weeks. In one woman, aged 37, the amenorrhœa had appeared after curettage for postpartum hæmorrhage; in the other, aged 29, it followed a difficult artificial labour with a severe puerperal infection. The child was stillborn.

Each woman keenly wished to have a child. Examination revealed a corpus uteri of normal size in a position of anteflexion. No pathological changes were observed in the adnexa. Intubation of the cervical canal was impossible; apparently it had been obliterated as far as the ostium externum.

Although there was no enlargement of the corpus uteri, the periodic pains suggested that a portion of the endometrium had been retained. Accordingly a new canal had to be created to restore the possibility of pregnancy. This was done by the following method:

Laparotomy: A median incision, extending from the portio vaginalis to the fundus, was made through the anterior wall of the uterus. A straight glass catheter, part of which had been cut off, was placed in the depth of the cleavage plane. The tip was fixed in the fundus with catgut. It was highly probable that the canal would be obliterated again after the catheter had been removed and this could only be prevented by rapidly covering it with epithelium. Since 1928 I have frequently

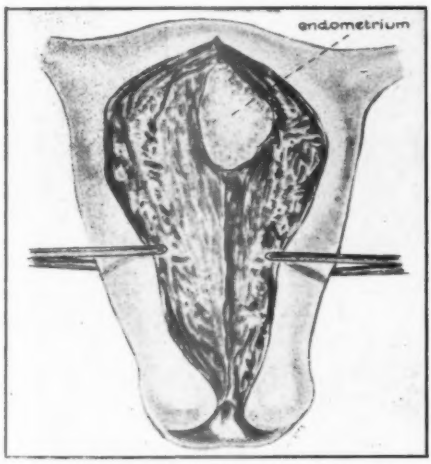

Fig. 1.—A longitudinal incision is made through the anterior wall of the uterus. There is a small cavity.

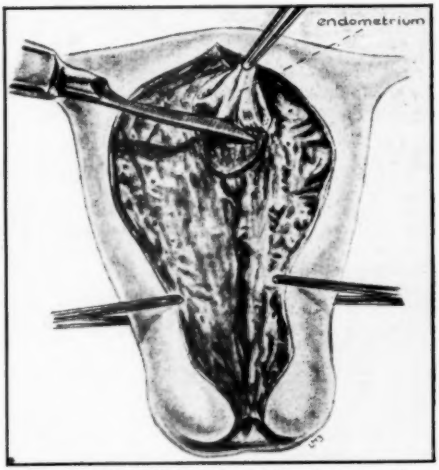

Fig. 2.—Part of the endometrium is removed.

inserted endometrium into the wounds of the excised cervix following a subtotal hysterectomy. Since then many others have also carried out this operation. Menstruation invariably recurred at the normal time, even when the implantation had been carried out at a time less than two weeks prior to the menstruation expected. It would therefore appear that the endometrium grows very rapidly in the tissue. Therefore I decided to remove a portion of mucosa from the uterine cavity still retained in our patients and to insert this around the glass catheter. The technique is shown in Figs. 1, 2, 3 and 4. Since the operation both women have menstruated normally without pain. I made a hysterosalpingography in both women: the oviducts were open in both cases. One of the women became pregnant again, the last menstruation occurring on 26.10.52. Unfortunately the patient no longer lived in Amsterdam, so that she was treated elsewhere. As she complained constantly of

pain in the lower abdomen, a cæsarean section was performed in the 34th week of pregnancy, for fear that a uterine rupture might occur. A child weighing 1,950 grammes was born, but unfortunately died within a few days from a hyaline-like membrane and absorption atelectasis. According to the attending obstetrician the entire anterior wall of the uterus was thin. However, there was no evidence of an imminent rupture. Although the wall of the uterus was thin, the obstetrician found the condition to be such that he believed he could recommend another pregnancy. So far the other patient has not become pregnant. Artificial insemination was attempted once but failed. Subsequently the woman has refused any further examination or treatment. In my opinion the absence of a mucus plug in the cervix must have considerably reduced the chance of pregnancy.

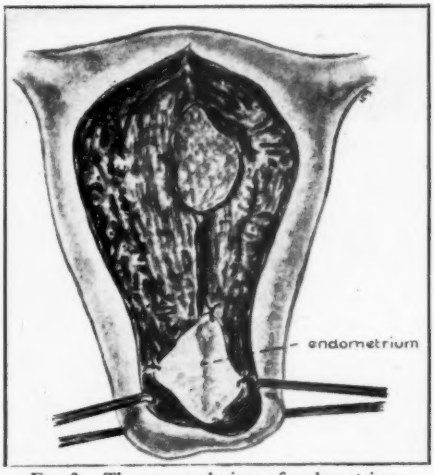

FIG. 3.—The removed piece of endometrium is sutured in the depth of the incision.

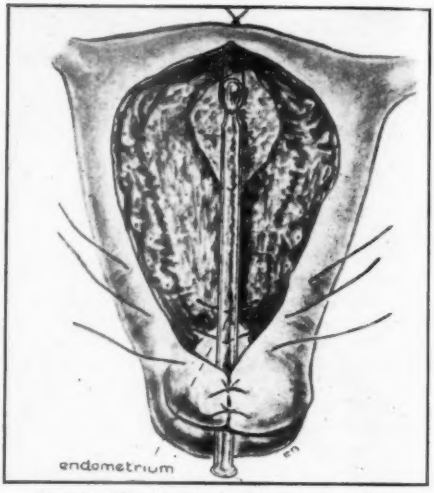

FIG. 4.—The incision is closed over a glass catheter.

The Treatment of Vesicovaginal Fistulas, Including So-called Radium Fistulas

By Professor M. A. VAN BOUWDIJK BASTIAANSE

Professor of Obstetrics and Gynæcology at the University of Amsterdam

IN treating vesical fistulas at the Obstetric Clinic of the University of Amsterdam the following points are taken into account:

(1) Prior to carrying out an operation, the site, the nature and the extent of the fistula is determined accurately.

(2) A sound is introduced into the urethra to make sure that it is not obliterated. This may occasionally occur.

(3) A vaginal approach is almost always to be preferred to an abdominal one. A combined abdominal and vaginal operation is hardly ever indicated, except in very large fistulas which are usually the result of radium therapy. I shall deal more fully with the treatment of these fistulas later.

(4) A good view and ready accessibility of the fistula is essential. This is obtained by marked flexion of the legs at the hip (Fig. 1) and a unilateral or bilateral episiotomy.

(5) The portion of the bladder wall surrounding the fistula should not be excised. This would merely enlarge the opening.

(6) The sutures must not be subject to tension. The bladder should be dissected out extensively, if possible so that the last sutures can be made in normal tissue. Suturing should be done at least in two, preferably in three, layers. This will result in the apposition of broad wound surface. Extensive dissecting out of the bladder involves the danger of making other openings. This danger may be avoided by grasping the margin of the vaginal wall directed toward the opening of the fistula with Kocher's forceps and using the latter to hold the vaginal wall tightly over the tips of the fingers. In this way the thickness of the tissue lying between the tip of the finger and the scissors, by which cutting is done, may be estimated correctly. By this method I have always been able to prevent the wall of the bladder from being injured again, even in the most difficult operations for fistulas.

(7) In the event of very extensive cicatrization it may sometimes be impossible to separate completely the bladder from the cicatricial tissue in all directions. A method must then be found of placing the movable part of the bladder wall over the closed opening in the form of a fold. The fold of the bladder wall may be sutured to the pelvic wall in some cases, far lateral to the vaginal wall in others, and sometimes, after the bladder has been dissected out, to the anterior wall of the uterus. When the suture is subjected to tension in an antero-posterior direction the anterior wall of the cervix may be sutured against or in the vicinity of the os pubis, after the vaginal wall has been dissected out at this point or the bladder pushed upward.

(8) In closing a fistula in which the urethra is involved as few vessels as possible should be ligated. Each ligation causes a local necrosis of tissue. Attention should be paid not only to the direction in which the ligatures are applied, but also to the course of the vessels, to ensure that as few as possible of these vessels are closed.

(9) No dead spaces must be left. Dead spaces may be prevented or filled by the following methods: (*a*) After the skin has been undermined, a pedicled fatty-tissue flap and bulbocavernosus muscle may be cut from the labium majus and inserted in the dead spaces (Martius' method). (*b*) When the dead space cannot be sufficiently filled by the flap a gynæcological tampon may be used to press the vaginal wall firmly against the subjacent tissue. However, a tamponade is rarely required.

FIG. 1.—Position for vaginal operation.

(10) When the posterior wall of the urethra is absent and the defect extends to within the bladder, a U-shaped incision is made and continued on the left and right side to just behind the clitoris. The edges of the wound are undermined. In addition, an incision is made through the anterior wall of the vagina extending from the lowermost part of the U-shaped incision to the cervix uteri. The vaginal wall is dissected out on both sides. A slender Nélaton catheter is inserted and the edges of the wound are sutured together around it. The tissue on either side of the orificium internum is sutured together in case it may still include functioning muscular tissue. The wall of the bladder is inverted by means of some transverse catgut sutures in order to reduce the tension on the suture of the closed fistula. The newly formed posterior wall of the urethra may become necrotic; therefore it is highly advisable to stitch a pedicled flap of adipose tissue, cut from one or both labia majora, to the suture, so that the flap extends from the orificium urethræ to the bladder. This method will almost certainly prevent a recurrence of the fistula.

To increase the effective function of the closing mechanism of the bladder, we usually proceed to carry out a sling operation with fascia strips of the rectus sheath.

(11) The suture material we use is catgut as silk or linen may give rise to calculi; nor should the catgut sutures be allowed to lie within the lumen of the bladder.

(12) When difficult or complicated operations of the bladder have to be carried out, the chance of recovery is increased considerably if the suture in the wall of the bladder is covered over either with peritoneum, an epiploic appendix, or with a slip of the omentum. The corpus uteri may also be used as plastic material. When the corpus uteri is absent, the bladder may be attached to the sigmoid at the site of the suture.

(13) A colpocleisis is indicated only in cases of enormous fistulas of the bladder and rectum especially when these have appeared after radium therapy. It is always possible to close the vesical and rectal fistula separately so that there is no longer any communication between the rectum and the bladder.

(14) There are no sharply defined surgical procedures to be used in all forms of vesical fistula. These must be determined in each individual case. Frequently various methods will have to be combined.

(15) When the bladder is injured in the course of an operation, this injury must be closed immediately.

A fistula may also be due to necrosis of tissue. An operation may only be performed when all necrotic tissue has been shed and the inflammation with which it is always accompanied has disappeared. This usually occurs within three to four months.

When an operation for fistula has failed one should also wait for three to four months before making another attempt.

Pre-operative treatment.—Infection of the genitals and of the surrounding skin should be prevented if possible. Sitz baths and irrigation with slightly disinfectant fluids may be employed. The state of the urinary tract is of great importance. Pyelitis must be treated. Fistulas are always associated with a more or less severe cystitis; when it is not severe no pre-operative therapy is required; when the cystitis is accompanied by a rise in temperature or pus in the urine, the operation must be postponed until the patient has recovered. We start with intramuscular administration of 400,000 units of penicillin daily one day prior to operation and continue this procedure for at least a few days post-operatively.

Prognosis

When the closure of the fistula has not been preceded by unsuccessful operations, prognosis is almost always favourable. When a previous attempt has failed, it will be less favourable. Each unsuccessful operation is followed by the appearance of cicatricial tissue, while the opening is usually enlarged as well.

Essential conditions for fistula operation are: (1) The site of operation should have normal tissue. (2) Adequate blood supply to the part. (3) Absence of tension at the sutures. (4) No infection in the operative field.

According to the literature the prognosis for extensive radium fistulas is very poor.

Post-operative treatment.—For an operation wound to heal properly the wound surfaces should always be immovably united with one another. Therefore the bladder must not be completely filled. This is prevented by using a catheter à demeure, left in position for eighteen to twenty-one days. However, an indwelling catheter has this drawback, it always acts as an irritant on the wall of the bladder and its use is frequently accompanied by the appearance of cystitis. This is not usually severe, however, and as a rule it has no effect on the healing of the wound. The intensity of the irritation will also depend upon the substance of which the catheter is made. Experience has shown that glass catheters are far less irritative than rubber catheters. We employ Skene's "horse-foot" catheter. When the urethra itself is involved in the fistula a slender indwelling Nélaton catheter is used.

The catheter must not become clogged. Rather no catheter than a clogged one. When there is no catheter the patient will be able to urinate spontaneously as soon as the bladder has been sufficiently filled. When a closed catheter is lying in the bladder this will often be impossible. Blocking of the catheter is mainly due to the precipitation of urates. To prevent this we irrigate the bladder every two hours with 10 to 20 ml. of fluid. This fluid consists of:

Citric acid	32·5 grammes
Magnesium oxide	3·84 grammes
Exsiccated sodium carbonate ..	4·37 grammes
Distilled water per 1,000 ml.	

Another precaution is to keep the urine acid. This will prevent the precipitation of phosphates. The nursing staff should see to it that the catheter runs freely. In my opinion it is advisable to leave the catheter in place for longer than the usual ten-day period. The catgut will have dissolved about the eighth day, and at this time there will be far more danger of distension as the result of an increased tension of the bladder than in the early post-operative period, when the catgut is still solid.

Sometimes the woman is advised to lie face downwards to prevent the urine from coming in contact with the suture. There never is a vacuum within the bladder, however, so the entire wall of the bladder will be in contact with the intravesical fluid and therefore it is useless to recommend a prone position. In our clinic all the patients lie in a lateral position to prevent the appearance of a thrombosis. We believe that pressure on the calf (dorsal position) causes closure of veins, resulting in anoxæmia, changes of the intima and thrombosis.

Finally, we urge our patients to take large quantities of fluid, as concentrated urine will be more liable to give rise to precipitation and therefore clogging of the catheter.

As far as the results are concerned, all the fistulas have closed anatomically. All patients with vesicovaginal fistulas, in whom the sphincter vesicæ was unimpaired, have also shown a functional recovery. The results of operations involving the sphincter vesicæ are shown in Table I.

TABLE I

	Number of cases	Anatomical and functional cure	Marked improvement
Vesicovaginal	37	37	
Urethro-vesicovaginal ..	33	26	6

In one case there was intense fibrosis around the urethra.

During the early years I did not employ the sling operation (Goebell-Stoeckel) as an additional routine procedure in the urethro-vesico-vaginal type of fistula. In the 7 cases in which this method was used 6 patients were cured and one was much improved.

The youngest patient on whom I have performed an operation combined with the Goebell-Stoeckel and the Martius' procedure for absence of the urethra with a defect extending up to and into the bladder, was 9 years of age. There was a complete recovery, both anatomically and functionally.

FISTULAS DUE TO RADIOTHERAPY

The prognosis as to the result of treatment of fistulas due to radiotherapy is far more unfavourable than that of fistulas due to other causes. Radium may cause tremendous destruction. We have operated on patients in whom the entire posterior wall of the bladder and a considerable part of the anterior wall of the rectum had disappeared (Fig. 2).

FIG. 2.—Condition before operation. The entire posterior wall of the bladder has disappeared.

In recent years 12 patients with radium fistulas of bladder and/or rectum were referred to us for treatment. Frequently it is impossible to dissect out the bladder to such an extent that the fistula can be sutured in a few layers without tension. The tissue is very stiff and the fibrosis involves all the tissues of the pelvis. In the case of a small fistula, the surrounding vaginal wall may be dissected out extensively, so that broad wound surfaces may be sutured together. When there is a large fistula, this will be impossible. So far I have, however, always been able to dissect out the bladder, or the rectum, or both, to such an extent that the edges, although subjected to tension, could be sutured together. For the wound to heal, tissue will also have to be applied to the suture as plastic material.

The best will be to use tissue lined with peritoneum. What type of tissue may be employed for this purpose?

(1) The corpus uteri: If the uterus is still present, the pouch of Douglas or the space between the bladder and the uterus may be opened and the corpus uteri drawn into the vagina, whereupon it may be sutured to the bladder or the rectum or, if necessary, to both. In the latter case hardly anything will be left of the vagina. Whenever I have used the uterus as plastic material, the cervix was scarcely movable owing to the marked fibrosis of the tissue. To prevent the uterus from being drawn back

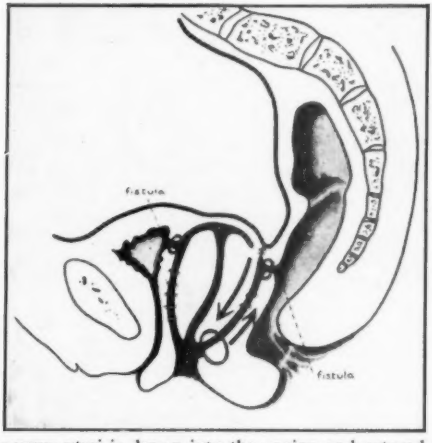

FIG. 3.—The corpus uteri is drawn into the vagina and sutured to the bladder and the rectum. The fundus of the uterus is perforated.

again over the suture, we have sutured the fundus to the levator muscles and in the tissue of the perineum with strong catgut. The skin of the perineum is then drawn forcefully inward. With this tissue the portion of the rectum situated distally to the fistula will also be drawn slightly upward, while the proximal portion will be drawn downward with the cervix uteri. This will slightly reduce the tension in the suture of the fistula (Fig. 3).

FIG. 4.

(2) Sigmoid and appendices epiploicæ: When the intestine is unimpaired, a fold of the sigmoid may sometimes be drawn slightly downward and stitched to the suture of the bladder.

(3) The fundus of the bladder and the anterior wall of the sigmoid: In the patients treated so far I have always been able to mobilize the fundus of the bladder and the anterior wall of the sigmoid to such an extent that they could be stitched as a fold over the suture (Figs. 4, 5, 6, 7). The fundus of the bladder may be mobilized by transverse cleavage of the peritoneum between the bladder and the anterior abdominal wall at laparotomy and by bluntly severing the bladder farther downward from the pubic bone. In addition the peritoneum and the subjacent tissue to the left and right of the

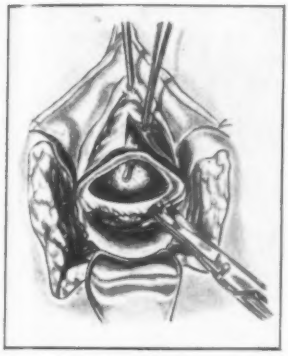

Fig. 5.

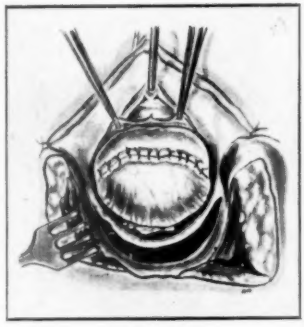

Fig. 6.

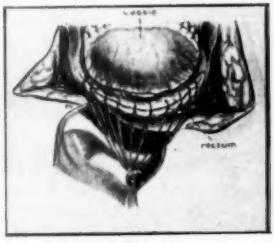

Fig. 7.

FIGS. 5, 6, and 7.—A fold of the fundus of the bladder is sutured over the first layer of sutures. The rectovaginal fistula is closed.

bladder must be divided. In view of the marked fibrosis of the tissue great caution should be observed with regard to the ureters. The anterior wall of the sigmoid may be mobilized by cleaving the peritoneum to the right and left of the intestine. Cleavage of the connective tissue below the sigmoid should be avoided, as the blood supply is considerably reduced in irradiated tissue, and the sigmoid may become necrosed.

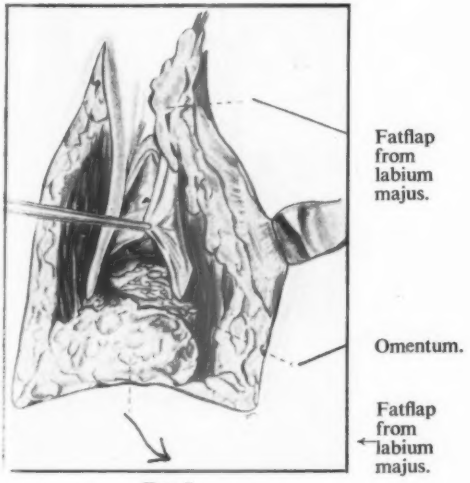

FIG. 8.

FIG. 9.—Final stage of operation.

(4) The omentum: The omentum always provides suitable tissue for suturing over intestinal sutures and for suturing over tissue deprived of peritoneum. The problem is to draw the omentum so far downward, that it may be introduced extensively into the vagina. This may be readily done when the omentum is divided transversely, parallel with the transverse colon, care being taken that some large vessels are still left to ensure the blood supply. To prevent ileus, the margin of the

omentum should be sutured laterally to the peritoneum of the anterior abdominal wall. In fistulas of the rectum a colostomy is performed a few weeks prior to operation.

We have often simultaneously employed several of the methods suggested, e.g. suturing ovc with the fundus of the bladder, with the sigmoid and with the omentum or with the uterus and omentum. When these methods are used, the vagina becomes obliterated. As the vagina will have in any case become a useless organ, it is advisable to fill its retained portion with pedicled adipose flaps cut from the labia majora (Martius' method), over which the vulva is then sutured. The prepared flaps of adipose tissue may be placed in position under the skin (Figs. 8 and 9). The greater the quantity of tissue filling the vagina, the greater the chance of closure of the fistula. Penicillin, streptomycin and, if necessary, a sulphonamide are applied to the wound cavity. Intramuscular injections of penicillin and, for a few days, of streptomycin as well, are given during the healing period.

In my opinion the greater part, even of the very largest radium fistulas, may be closed by the method described. Among the 12 patients with radium fistulas on whom I have operated there was only one, a 78-year-old woman, in whom a small vesical fistula persisted (Table II).

TABLE II.—RESULTS OF SURGICAL TREATMENT OF FISTULAS DUE TO RADIUM TREATMENT

	Number	Anatomical and functional cure
Vesicovaginal	2	2
Vesico-rectovaginal	7	6*
Rectovaginal	3	3

*In one case a small fistula persisted.

Initially I believed that in very large fistulas, the bladder and the rectum might be separated more readily from the abdomen than from the vagina. This has proved incorrect. When proceeding from the abdomen, the boundary between the bladder and the rectum can no longer be determined. Nor can this be done when the tip of the finger is introduced into the vagina by an assistant.

I have not come across the method I have described in the literature. This procedure may be regarded as a useful addition to the methods used to close fistulas due to radium therapy.